VNIVERSITAT DE VALÈNCIA

Facultad de Psicología

Programa de Doctorado 3153

"Psicología y Psicopatología Perinatal e Infantil"

TESIS DOCTORAL CON MENCIÓN INTERNACIONAL

"Social withdrawal behavior in infants with cleft lip and palate: the psychological impact of the malformation on parents and on parents-infant relationship"

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Valencia, diciembre de 2017.

Programa de Doctorado 3153 en Psicología y Psicopatología Perinatal e Infantil (Universitat de València, España), regulado por RD 99/2011.

A mis padres

Maricela y Carlos

ACKNOWLEDGEMENTS

A través de estas líneas quisiera agradecer a todas las personas que me acompañaron y apoyaron en este proceso, quienes más allá del aspecto académico, me han transmitido muchas enseñanzas a nivel personal.

Cualquier agradecimiento es poco para mis padres, Maricela y Carlos, quienes me han motivado desde mi infancia para alcanzar lo que parecía inalcanzable. Gracias por su dedicación, por su apoyo, por su ejemplo.

Gracias infinitas a Toni, por recorrer juntos este camino. Por ser mi inspiración cada día, por creer en mí y por ser el mejor compañero de vida que pude encontrar.

A Amparo y Andrés, quienes siempre tienen la palabra adecuada para seguir adelante. Gracias por sus consejos, por su cariño, por su gran ejemplo en la familia y en el trabajo.

Gracias a Sebastián, por ser tan adorable.

A mis directores de tesis, Antoine Guedeney, Dolores Frías Navarro y Sandra Simó Teufel. A todo el equipo de FENTES que ha hecho posible este trabajo. En ESTRASBURGO: Grollemund Bruno, Idrissi Hanan, Hecketsweiler Stéphane, Bruant-Rodier Catherine, Kauffmann Isabelle, Danion-Grilliat Anne, Corduan Guillaume, Velten Michel, Hamadi Abdelkader, Quentel Elvire, Boudas Raphael; en LILLE: Pellerin Philippe, Frochisse Caroline, Martinot Duquesnoy Véronique, Delpierre Vanessa, P. Delion, Kosmala Aurélie; en PARÍS: Vazquez Marie-Paule, Soupre Véronique, Galliani Eva, Zazurca Frédéric, Gavelle Pascalle, Brute Michèle, Maillart Aurélie, Guedeney Antoine, Dubois de Bodinat Ludivine; en NANCY: Etienne Simon, Davrou Julien, Kabuth Bernard, Gall Jean-Yves, Forter Catherine. En especial, a Bruno Grollemund y a Pascale Gavelle por su disponibilidad y su amabilidad en el trabajo y fuera de este. *Je vous remercie de m'avoir permis de faire partie de cette recherche.*

Especialmente, quisiera agradecer a todas las familias que participaron en este proyecto de manera voluntaria. Gracias por permitirnos aprender de su experiencia.

Quisiera agradecer también a quienes hicieron posible mis estancias de investigación en París y en Bogotá. A Pascual Palau por su apoyo. A mi *Alma Mater*, la Universidad Nacional Autónoma de México, gracias a cuyas enseñanzas he podido avanzar en esta etapa. Finalmente, quisiera mencionar que este trabajo fue posible gracias a la beca otorgada por el Consejo Nacional de Ciencia y Tecnología de México (CONACYT) y el Consejo Oaxaqueño de Ciencia y Tecnología (COCYT) para realizar mis estudios de Doctorado en la Universidad de Valencia.

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RESUMEN

La malformación de labio y paladar hendido (CL/P) es una de las malformaciones cráneo-faciales más frecuentes en humanos. Aunque esta condición médica no es una de las principales causas de mortalidad en los países desarrollados, conlleva una considerable morbilidad al paciente e impone una carga sustancial sobre la salud, la calidad de vida y el bienestar socioeconómico de los infantes y sus familias.

En la actualidad, ningún protocolo genérico ha sido reconocido por toda la comunidad médica para el tratamiento de los infantes con CL/P y sus familias. Por tanto, cada paciente y su familia son atendidos dependiendo de la experiencia y la toma de decisiones del equipo médico. Además, el apoyo psicológico se ofrece sólo en algunos centros de referencia, y existe una falta de conocimiento sobre el estado psicológico de los infantes con CL/P y sus familias durante el primer año de vida. Por consiguiente, resulta necesario realizar investigaciones que permitan la identificación de indicadores de riesgo para el bienestar psicológico en los infantes con CL/P y sus padres con la finalidad de proveer un apoyo psicológico adecuado.

En este sentido, la presente tesis doctoral tiene como objetivo general describir la evolución del retraimiento social infantil, el impacto psicológico en los padres y la calidad de la relación padres-bebé, a los cuatro y a los doce meses postparto, en infantes con labio y paladar hendido (CL/P) y sus padres. De manera específica, se exploró, a los cuatro y a los doce meses, las posibles diferencias en estas variables entre los grupos según el tipo de malformación (CLP: Labio y Paladar Hendido o CL: Labio Hendido), la lateralidad de la malformación (unilateral o bilateral), el tipo de diagnóstico (prenatal o postnatal) y el momento de la primera cirugía de reparación (temprana o tardía).

La muestra total del estudio consistió en 145 infantes y sus padres en la primera evaluación, y 123 infantes y sus padres en la segunda evaluación. Los bebés tenían una edad promedio de 4.04 meses (*DE* = 0.65) en la primera evaluación, y 12.29 meses (*DE* = 1.24) en la segunda evaluación. El 69.7% de los infantes participantes eran varones, y el 40.1% de los casos eran primogénitos. Por su parte, las madres tenían una edad promedio de 30.82 años (*DE* = 5.13), y los padres de 33.38 años (*DE* = 5.88). Todos los bebés con labio y paladar hendido (CL/P), nacidos y tratados en los centros participantes, fueron elegibles para formar parte de este estudio. Los criterios de no inclusión fueron: lactantes nacidos antes de las 34 semanas de gestación, bebés cuyo peso al nacer fuera inferior a 2.00 kg, que estuvieran en hogares de acogida, o cuyos padres fueran analfabetos o tuvieran conocimientos insuficientes de francés.

De la muestra total, el 80.3% de los infantes tenían labio y paladar hendido (CLP), y el 19.7% restante presentó sólo labio hendido (CL). De acuerdo a la lateralidad de la malformación, el 79.5% tuvo una malformación unilateral, y el 20.5% fue bilateral. El 84.2% de los padres recibió el diagnóstico de la malformación durante el periodo prenatal, y el 43.8% de los infantes tuvo cirugía de reparación labial durante los primeros 90 días postparto (temprana).

La metodología de este estudio fue no experimental y el diseño fue prospectivo, descriptivo y longitudinal. Se llevaron a cabo dos evaluaciones, la primera a los cuatro meses (T0) y la segunda a los doce meses postparto (T1). En ambas evaluaciones se utilizaron los mismos instrumentos de medición. Para evaluar el comportamiento de retraimiento infantil se utilizó la escala ADBB (*Alarm Distress Baby Scale)* (Guedeney & Fermanian, 2001) y la versión corta m-ADBB (Matthey, Crncec, Hales, & Guedeney, 2013).

La calidad de la relación padres-bebé se evaluó a través de la escala PIPE (*Pediatric Infant Parent Exam)* (Fiese, Poehlmann, Irwin, Gordon, & Curry-Bleggi, 2001). El impacto psicológico en los padres se operacionalizó a través de la evaluación del impacto en la familia (Impact On Family Scale) (Boudas, Jégu, Grollemund, Quentel, Danion-Grilliat, & Velten, 2013), el malestar psicológico (*Psychological Distress Index)* (Préville, Boyer, Potvin, Perrault, & Légaré, 1992), la depresión postnatal *(Edinburgh Postnatal Depression Scale)* (Guedeney & Fermanian, 1998) y la satisfacción marital *(Dyadic Adjustment Scale*) (Antoine, Christophe, & Nandrino, 2008). En nuestro estudio, todos los instrumentos de medición obtuvieron una consistencia interna buena o aceptable (alfa de Cronbach de .65 a .91), lo que garantiza su uso en infantes con CL/P y sus familias.

Con respecto al análisis de los datos, se llevaron a cabo análisis descriptivos de los ítems y de las puntuaciones totales de cada instrumento de medición. Posteriormente, con el fin de explorar la evolución -entre la primera y la segunda evaluación- del retraimiento social infantil, el impacto psicológico en los padres y la calidad de la relación padres-bebé, se realizó un análisis de varianza (ANOVA) para medidas repetidas (intra-sujetos). Además, para explorar las diferencias entre el tipo de malformación, la lateralidad de la malformación, el tipo de diagnóstico y el momento de la primera cirugía de reparación, se realizó un análisis de varianza (ANOVA) entre grupos. Todos los análisis se realizaron con el software estadístico SPSS, con intervalos de confianza del 95%.

Los resultados indicaron que, de manera general, el retraimiento social infantil, el impacto en la familia, el malestar psicológico paterno y la depresión posparto paterna fueron significativamente mayores a los cuatro meses (T0), en comparación a los doce meses (T1). Es decir, en estas variables hubo una evolución positiva hacia la adaptación a la

condición médica por parte de los infantes y los padres, durante el primer año.

En este sentido, los infantes mostraron una evolución positiva en términos de un menor nivel y porcentaje de retraimiento social en la segunda evaluación, en comparación con la primera medición. A los cuatro meses, el 15.9% de los infantes presentó signos de retraimiento social –de acuerdo con la escala ADBB- y el 24.9% según la escala m-ADBB. A los doce meses, estos porcentajes disminuyeron al 10.6% –según la escala ADBB- y al 13.8% de acuerdo con la escala m-ADBB. Por otra parte, los padres reportaron significativamente menores niveles de malestar psicológico y menores síntomas depresivos a los doce meses, que a los cuatro meses. Es decir, los padres mostraron una evolución positiva durante el primer año de vida de su hijo/a con CL/P.

En cuanto al tipo de malformación, a los cuatro meses, se observó un mayor impacto en la familia -reportados por madres y padres- y un menor nivel de satisfacción marital materna en el grupo de CLP, en comparación con el grupo de CL. De manera similar, a los doce meses, las madres del grupo CLP reportaron un mayor impacto en la familia, en comparación con el grupo de CL. Es decir, en ambas evaluaciones, cuando la malformación infantil era más compleja (CLP), el impacto psicológico en los padres fue mayor.

Según la lateralidad de la malformación, a los cuatro meses se observó que la satisfacción marital en los padres fue significativamente mayor en el grupo de malformación bilateral. Sin embargo, a los doce meses no se encontraron diferencias estadísticamente significativas entre los grupos. Por tanto, podría decirse que la lateralidad de la malformación tiene escasa relevancia en el impacto psicológico en el infante con CL/P y sus padres.

En lo que respecta al tipo del diagnóstico, en la primera evaluación el impacto en las madres y los padres fue significativamente mayor en el grupo de diagnóstico prenatal, en comparación con el de diagnóstico postnatal. De manera interesante, en la segunda evaluación la calidad de la relación padres-bebé fue significativamente mayor en el grupo de diagnóstico prenatal. Cabe señalar que no se observaron diferencias en el retraimiento social infantil entre los grupos de diagnóstico. Estos resultados indicarían que los padres que recibieron un diagnóstico prenatal (vs. diagnóstico postnatal) suelen estar más afectados durante los primeros meses posteriores al nacimiento de un hijo/a con CL/P. Sin embargo, a largo plazo, el diagnóstico prenatal podría constituir un factor protector para la relación padres-bebé.

Finalmente, de acuerdo al tiempo de espera para la primera cirugía de reparación, se observó que en la primera evaluación el impacto en la familia fue significativamente mayor, y las madres mostraron mayores síntomas depresivos cuando la cirugía de reparación labial se realizó después de los 90 días postparto (tardía). En la segunda evaluación el impacto en la familia, la depresión postparto –reportados por ambos padres-, así como el malestar psicológico paterno fueron significativamente mayores en el grupo de cirugía tardía.

Estos hallazgos señalarían que, a mayor tiempo de espera para la primera cirugía de reparación, mayor es el impacto psicológico en los padres. Por tanto, una cirugía de reconstrucción dentro de los primeros 90 días postparto reduciría el impacto psicológico en los padres. Cabe mencionar que en el presente estudio no se observaron diferencias significativas en el retraimiento social infantil, ni en la calidad de la relación padres-bebé entre los grupos de cirugía temprana y tardía. Es decir, el tiempo de espera para la primera cirugía parece no estar ligado al nivel de retraimiento social infantil o a la calidad de la relación padres- bebé, pero sí al impacto psicológico en los padres.

En resumen, los resultados de la presente investigación mostraron un impacto psicológico significativamente mayor en los bebés y en los padres –en términos de retraimiento social infantil, impacto en la familia, malestar psicológico paterno y síntomas depresivos paternos- en la primera evaluación, en comparación con la segunda. El hecho de que el retraimiento social infantil haya disminuido significativamente a lo largo del primer año podría deberse a que en los primeros meses de vida el bebé está sujeto a diversos tratamientos médicos intrusivos, por ejemplo, para facilitar la alimentación, lo que aumenta el riesgo de desarrollar retraimiento social. Aunado a lo anterior, a los doce meses de vida el infante cuenta con una mayor cantidad y variedad de estrategias para afrontar el estrés y el malestar somato-emocional, más allá del retraimiento social.

Asimismo, la disminución del impacto psicológico en los padres puede explicarse mediante el modelo de estrés familiar y de afrontamiento (McCubbin, Thompson, & McCubbin, 1996), el cual establece que las familias con un bebé con una condición médica buscan restablecer el equilibrio, con el fin de progresar a un estado de adaptación a largo plazo. En esta línea, los padres mostraron una evolución más positiva en la adaptación a la malformación de sus hijos/as, mostrando niveles significativamente menores de impacto psicológico a los doce meses.

En relación a las diferencias entre los grupos, se observó que cuando el infante presentaba una malformación más compleja (CLP), los padres reportaban un nivel más elevado de impacto psicológico. Estos resultados indicarían una mayor necesidad de apoyo psicológico en familias con un infante con CLP –en comparación con el grupo de CL-, durante el primer año, para reducir el malestar emocional y favorecer la adaptación de la familia a la condición médica del bebé. Por otro lado, se observó que la lateralidad de la malformación es una variable con escasa relevancia con respecto al bienestar psicológico de los infantes, de los padres o de la relación padres-bebé.

Por su parte, el tipo de diagnóstico fue un factor clave en el impacto psicológico en los padres y en la relación padres-bebé. En el grupo de diagnóstico prenatal, se hizo evidente un mayor impacto psicológico en los padres, en los primeros meses de vida del bebé. A largo plazo, se observó una menor calidad de la relación padres-bebé en el grupo de diagnóstico postnatal. Estos hallazgos destacan la importancia de ofrecer información sobre el origen y el tratamiento de la malformación, y de dar apoyo psicológico a los padres desde el momento del diagnóstico –incluso desde la etapa prenatal- para reducir el nivel de malestar psicológico parental y para favorecer estilos de interacción más adaptativos.

Uno de los resultados más importantes de esta investigación fue que una cirugía de reparación labial temprana (realizada durante los primeros 90 días de vida del bebé) contribuye a preservar el estado psicológico de los padres durante el primer año. Esta información es especialmente útil para los equipos médicos quirúrgicos, pues señalaría una gran ventaja de las intervenciones tempranas sobre las cirugías tardías. En este sentido, estos resultados aportan una valiosa información, desde una perspectiva psicológica, para facilitar el consenso de un protocolo médico quirúrgico que favorezca las cirugías de reparación tempranas.

Además, este es el primer estudio que proporciona información sobre la conducta de retraimiento social en infantes con CL/P durante el primer año de vida. Cabe mencionar que, aunque no se encontraron diferencias en el retraimiento social infantil entre los diferentes grupos de tipo y lateralidad de la malformación, el tipo de diagnóstico y el momento de la cirugía de reparación, los porcentajes reportados a los cuatro y a los doce meses corresponden a los de poblaciones en riesgo. Por tanto, la evaluación del retraimiento social, como signo de sufrimiento emocional infantil, resulta especialmente necesaria en

infantes con una condición médica –como la malformación de CL/P- durante los primeros meses de vida.

Otra importante contribución de este estudio fue la adopción de un enfoque integral para conocer el ajuste y la evolución de los padres y el bebé ante la malformación CL/P. Hasta el momento, la mayoría de las investigaciones se ha centrado en el estudio de la diada madre-bebé, dejando de lado la contribución del padre en el bienestar psicológico de la familia. Por esta razón, en nuestro estudio se pretende conocer la forma en la que los padres hacen frente a las situaciones estresantes familiares como lo es la condición médica de un hijo/a.

En conclusión, se observó que los padres y los infantes que participaron en nuestro estudio mostraron una buena adaptación a la condición de la malformación de CL/P, ya que el nivel de retraimiento social y el impacto psicológico en los padres fue menor en la segunda evaluación, en comparación con la primera medición. Además, esta adaptación fue más positiva cuando el infante presentaba una malformación menos severa (Labio Hendido), cuando el diagnóstico se conocía desde la etapa prenatal, y cuando la cirugía de reparación se realizaba tempranamente.

Los hallazgos de nuestra investigación proporcionan indicadores que permiten la identificación de familias en alto riesgo de presentar problemas de salud mental, y proveen directrices para la prevención y la intervención psicológica temprana. Además, nuestros resultados enfatizan la necesidad de incluir a profesionales de la salud mental como parte del equipo interdisciplinario que atiende a las familias con un infante con la malformación CL/P. Ya que a través del apoyo psicológico especializado se facilitaría la transición de los padres y los infantes hacia la adaptación y el afrontamiento de los diversos factores estresantes asociados a la malformación de labio y paladar hendido.

Palabras clave: retraimiento social infantil, labio y paladar hendido, malformación, impacto psicológico en los padres, relación padres-bebé, cirugía de reparación, diagnóstico prenatal y postnatal.

ABSTRACT

Cleft lip and palate (CL/P) malformation is one of the most frequent craniofacial malformations in humans. This medical condition is not a major cause of mortality in developed countries, although, it causes considerable morbidity to the patient and imposes a substantial burden on the health, quality of life and socio-economic wellbeing of infants and their families. Nowadays, no generic protocol has been recognized by the entire medical community for caring infants with CL/P and their families. Hence, each patient and his/her family is cared depending on the experience of the medical team. Moreover, psychological support is offer only in the expert centers, and there is a lack of knowledge about the psychological state of the infants with CL/P and their families during the first year postpartum. Therefore, it is necessary further research that allows the identification of indicators in families with higher risk for psychological distress in order to implement appropriate support to infants with CL/P and their parents.

Accordingly, this thesis has as general objective to describe the evolution of the infant social withdrawal behavior, the psychological impact on parents, and the quality of the parent-infant relationship, at four and at twelve months postpartum, in infants with Cleft Lip and Palate malformation. The specific goals were to explore the differences in these variables between the groups according to the type of malformation (CLP: Cleft Lip and Palate or CL: Cleft Lip only), the laterality of the malformation (unilateral or bilateral), the type of diagnosis (prenatal or postnatal) and the timing of the repair surgery (early or late).

The total sample of the study consisted of 145 French infants and their parents in the first assessment, and 123 infants and their parents in the second assessment. Infants were an average age of 4.04 months (*SD* = 0.65) at the first evaluation, and 12.29 months (*SD* = 1.24) at

the second assessment. Of the total sample, 69.7% of the infants were male, and 40.1% were the first born. Moreover, mothers had an average age of 30.82 years (*SD* = 5.13), and fathers 33.38 years (*SD* = 5.88). All infants born with a cleft lip (with or without cleft palate) and followed in the recruitment centers were eligible for the study. Non-inclusion criteria were infants born before 34 weeks of gestation, infants whose birth weight was under 2.00 kg, placed in foster homes, and whose parents were illiterate or showed insufficient level of French.

According to the type of malformation, 80.3% of the infants had Cleft Lip and Palate (CLP), thus, 19.7% presented with a Cleft Lip only (CL). Of the total sample, 79.5% had a unilateral malformation; 84.2% of the parents received the diagnosis of their infant's malformation prenatally; and 43.8% of the infants had the lip repair surgery during the first 90 days postpartum.

The methodology of this study was non-experimental and the design was prospective, descriptive, and longitudinal. Two assessment sessions were carried out, one at four months (T0) and the other at twelve months postpartum (T1). In both measurements, the same evaluation instruments were used. The ADBB scale (Alarm Distress Baby Scale) (Guedeney & Fermanian, 2001) and the short version m-ADBB scale (Matthey, Crncec, Hales, & Guedeney, 2013) were used to assess infant social withdrawal behavior. The quality of the parent-infant relationship was assessed through the Pediatric Infant Parent Exam (PIPE) (Fiese, Poehlmann, Irwin, Gordon, & Curry-Bleggi, 2001).

The psychological impact on parents was operationalized through the impact on family (Impact On Family Scale) (Boudas, Jégu, Grollemund, Quentel, Danion-Grilliat, & Velten, 2013), the psychological distress on parents (Psychological Distress Index) (Préville, Boyer, Potvin, Perrault, & Légaré, 1992), the postnatal depression on parents

(Edinburgh Postnatal Depression Scale) (Guedeney & Fermanian, 1998) and the marital satisfaction (Dyadic Adjustment Scale) (Antoine, Christophe, & Nandrino, 2008) on the parental couple. All instruments used in the present research reached a good or acceptable internal consistency (Cronbach's alpha from .65 to .91), which guaranties their use in infants with CL/P and their families.

Regarding the analysis of the data, descriptive analysis of the items and the total scores of each instrument was carried out. Subsequently, in order to explore the evolution -between the first and the second evaluation- of the infant social withdrawal behavior, the psychological impact on parents and the quality of parent-infant relationship, we carried out an analysis of variance (ANOVA) for repeated measures (within subjects). Besides, to explore the differences between the groups in terms of the type of malformation, the laterality of the malformation, the type of diagnosis and the timing of the repair surgery, we performed an analysis of variance (ANOVA) between groups. All the analyses were performed with the SPSS statistical software, with 95% confidence intervals.

In general, results indicate that the infant social withdrawal behavior, the impact of the malformation on the family, as well as paternal psychological distress and postpartum depression in fathers were significantly higher at four months (T0) than at twelve months (T1) postpartum. That is, infants showed a positive evolution in terms of social withdrawal behavior between the first and the second assessment. At four months 15.9% of the infants were socially withdrawn according to the ADBB scale, while 24.9% were identified as withdrawn with the m-ADBB scale. At twelve months, 10.6% of the infants showed signs of social withdrawal behavior -assessed by ADBB scale- and 13.8% were identified as withdrawn with the m-ADBB scale.

Mothers only showed a significantly reduce in psychological impact. And fathers reported significantly less psychological distress and depressive symptoms at twelve months than at four months. That is, fathers showed a good evolution during the first year after the birth of their infant's having CL/P.

Concerning the type of malformation, at four months, parents of the Cleft Lip and Palate group (CLP) showed significantly higher levels of impact on family –reported by mothers and fathers- and significantly lower maternal marital satisfaction, comparing with the Cleft Lip group (CL). No differences were found in infant's social withdrawal either in quality of the parent-infant relationship between CLP and CL groups in the first assessment. At twelve months, only the impact on family reported by mothers was significantly higher in the CLP group, in comparison to the CL group. Thus, in both evaluations, when the infant's malformation was more severe (CLP), the psychological impact on parents was higher.

Regarding the laterality of the malformation, at four months, only marital satisfaction on fathers was significantly higher in the group with bilateral malformation. Nevertheless, at twelve months, no significant differences were observed. Therefore, it could be said that the laterality of the malformation has little relevance in the psychological impact of CL/P malformation in the infants and their parents.

In regard the timing of the diagnosis, in the first assessment, the impact on family –reported by mothers and fathers- was significantly higher in the group of prenatal diagnosis, compared to the postnatal diagnosis. Interestingly, in the second evaluation, the quality of parent-infant relationship was significantly higher in the group of prenatal diagnosis. No differences in infant's social withdrawal between the prenatal and the postnatal diagnosis were observed. These results would indicate that during the first months after the birth of an infant

with CL/P, parents who received a prenatal diagnosis (vs. postnatal diagnosis) are usually more affected. However, in the long term, prenatal diagnosis may be a protective factor for the parent-infant relationship.

Finally, it was observed that, at four months postpartum, the impact on family –reported by mothers and fathers- was significantly higher, and mothers showed significantly more depressive symptoms when the infant had a late lip repair surgery. At twelve months postpartum, impact on family –reported by both parents-, psychological distress in mothers and fathers, and paternal depressive symptoms were significantly higher in the late surgery group. However, no significant differences were observed in infant's social withdrawal or in quality of parent-infant relationship between the early and the late surgery groups.

In general, our results showed a significantly higher psychological impact –in terms of infant social withdrawal behavior, impact on family, psychological distress and depressive symptoms on fathers- at four months postpartum, compared to twelve months. The fact that infant social withdrawal behavior had decreased significantly over the first year may be because in the first months of life the baby receives various intrusive medical treatments, for example to facilitate feeding, which increases the risk of developing social withdrawal. Likewise, at twelve months, the infant has a greater quantity and variety of strategies to cope with emotional distress, beyond social withdrawal behavior.

Similarly, the diminution of the psychological impact on parents may be explained by the family stress and coping model (McCubbin, Thompson, & McCubbin, 1996), which states that families having an infant with a medical condition seek to restore balance and progress to a state of long-term adaptation. In this line, fathers showed a good

evolution in psychological impact of the malformation during the first year postpartum. That is, fathers showed less difficulties in adaptation to their infant's CL/P malformation.

Furthermore, it was demonstrated that parents having an infant with CLP showed higher levels of psychological impact compared to parents having an infant with a single CL malformation. This emphasized the greater need of psychological support to parents having an infant with a more severe malformation, during the first year postpartum. We also found that the laterality of the malformation does not necessarily influence the wellbeing of infants, parents, or the quality of the parent-infant relationship.

However, the timing of the diagnosis was a key factor to consider because we found that parents of infants diagnosed prenatally had higher impact on family, at four months evaluation. At twelve months, it was observed that prenatal diagnosis was a long-term protective factor for the quality of parent-infant relationship. These results highlight the importance of support and information about the CL/P malformation, which should be given as soon as possible, in order to reduce the psychological concern on parents and to facilitate more adaptive interaction patterns.

One of the most important contributions of the current study was that an early lip repair surgery (performed during the first 90 days of infant's life) significantly contributes to preserving the parental psychological wellbeing during the first year postpartum. This information is especially useful for surgical medical teams, as it indicates a great advantage of early versus late repair surgeries. In this line, these results provide valuable information, from a psychological perspective, to facilitate the consensus of a surgical medical protocol in favor of early repair surgeries.

Besides, this is the first study which provides information about social withdrawal behavior in infants with CL/P during the first year postpartum, emphasizing the importance of screening this sign of emotional distress in infants during the early postpartum period, in particular in the context of CL/P malformation. Moreover, another contribution of this study was the adoption of a comprehensive approach to parental and infant adjustment to CL/P malformation, taking into account the father's perspective, and using different dimensions of parent's mental health (impact of the malformation on the family life, psychological distress, depression, and marital satisfaction).

In conclusion, it was observed that parents and infants showed a good adaptation to the condition of the CL/P malformation, since the level of infant social withdrawal behavior and the psychological impact on parents was lower in the second assessment. In addition, this adaptation was more positive when the infant had a less severe malformation (Cleft Lip only), when the diagnosis was known from the prenatal stage, and when the repair surgery was performed early.

Finally, our findings provide indicators that allow the identification of families at high risk for mental health problems and provide guidelines for prevention and early psychological intervention. Additionally, our results emphasize the need to have a mental health professional as part of the interdisciplinary team that cares for families with an infant with CL/P malformation. Such specialized psychological support would facilitate the transition of parents and infants towards the adaptation and coping of the various stressors associated with CL/P malformation.

Key words: infant social withdrawal behavior, cleft lip and palate, psychological impact on parents, parent-infant relationship, repair surgery, prenatal and postnatal diagnosis.

PRESENTATION

The need for knowledge that arises in this doctoral thesis is to describe the evolution of the infant social withdrawal behavior, the psychological impact on parents, and the quality of the parent-infant relationship in infants with Cleft Lip and Palate (CL/P) and their parents, at four and at twelve months postpartum. Additionally, we aim to explore the differences in these variables between the groups of type of the malformation, the laterality of the malformation, the type of diagnosis performed, and the waiting time prior to the first surgery.

Cleft lip with or without palate (CL/P) is one of the most frequent craniofacial malformations in humans. The disruption of normal facial structure is immediately recognizable and this may have important consequences on parents' perception of their infant. For instance, it has been shown that infants perceived as less attractive receive routine caregiving, in comparison to those perceived as more attractive who more often receive affectionate care from their parents (Langlois, Ritter, Casey, & Sawin, 1995). Furthermore, the disfigurement makes infants with clefts (CL/P) less appealing (Goodacre, Henteges, Moss, Short, & Murray, 2004) and it is more difficult for parents to interpret their expressions (Field & Vega-Lahr, 1984).

All these elements are crucial in order to develop an adaptive parent-infant relationship and to facilitate infant mental wellbeing. Additionally, it has been showed that infants with CL/P made fewer communicative signals toward their mothers, producing fewer positive vocalizations, spent less time in visual contact with their mothers, were less engaged in active exploring of the environment and exhibited more self-absorbed behavior (Montirosso et al., 2012; Murray et al., 2008). All these behaviors are signs of social withdrawal, which may be interpreted as psychological distress in the infant. Accordingly, in the current study, we aimed to describe the evolution of the social

withdrawal behavior in infants with cleft lip and palate (CL/P), through the evaluation with the ADBB scale at four and at twelve months postpartum.

In the context of a medical condition, psychological difficulties are not just limited to infants, but also to their parents. It has been observed that parents of infants with CL/P may experience an initial emotional reaction of shock, confusion, sadness, anger, and guilt (Bradbury & Hewison, 1994), before they are able to form a bond with their babies (Drotar, Baskiewicz, Irvin, Kennell, & Klaus, 1975). Considering that infants can recognize adult emotional states and respond to them (Cohn & Tronick, 1983), parental emotional statements can be expected to affect the early interaction between parent and infant, and thus the infant's further development.

Moreover, the presence of CL/P in infants, as the impairment affects facial expressiveness and vocal production, may complicate the mother's interpretation of infant signals, and, in turn, this behavior might lead to less infant involvement in the interaction and more social withdrawal (Montirosso et al., 2012). In addition, the disfigurement of the infant with CL/P may influence the emotional wellbeing of their parents, and this psychological impact may negatively affect development (e.g., Turner, Rumsey, & Sandy, 1998).

Previous studies have demonstrated that the quality of parent-infant interaction differs significantly between dyads with a cleft lip (with or without cleft palate), compared with control dyads (Barden, Ford, Jensen, Rogers-Salyer, & Salyer, 1989; Field & Vega-Lahr, 1984; Koomen & Hoeksma, 1992; Langlois, Ritter, Casey, & Sawin, 1995; Montirosso et al., 2012; Speltz, Goodell, Endriga, & Clarren, 1994; Wasserman, Allen, & Solomon, 1985; Wasserman, Allen, & Linares, 1988). However, almost all of the literature that currently exists on this theme has focused on the mothers, resulting in a lack of research

involving the fathers of infants with cleft lip and palate. On the basis of these findings, the present research aimed also to evaluate the psychological impact on both mothers and fathers, as well as the quality of parent-infant interactions, at four and at twelve moths postpartum.

Regarding the several interventions and treatments, currently, no generic protocol has been recognized by the entire medical community. Each patient is cared for according to the experience and the decisions of the medical team in charge. Thus, the attention given to the psychological state of the infant and their parents vary depending of the team. Only the expert centers have specifically committed psychologists. But nowhere is the psychological care of infants with CL/P and their parents clearly protocoled, as is the case almost everywhere in France, for example in the case of prematurity. This is quite strange considering the frequency of the CL/P malformation, as it can be considered a *cas d'école* for Liaison Psychology in perinatality.

Furthermore, it is also necessary to investigate how early diagnosis can affect parents and the parent-infant interaction, even when this diagnosis is confirmed after birth. It has been suggested that there is an urgent necessity to assess the timing of repair surgeries to determine if earlier interventions optimize the infant's development (Yazdy, Honein, Rasmussen, & Frias, 2007), and how to reduce the psychological impact on parents and on the parent-infant relationship, which is clearly a major issue.

In this line, a recent review of the literature has emphasized the ongoing need for implementation of appropriate support in order to achieve psychological wellbeing and positive adjustment for families with an infant with CL/P (Nelson, Glenny, Kirk, & Caress, 2012). Some surgeons (e.g., Rey-Bellet & Hohlfeld, 2004) have developed the habit of going to the Gynecology and Obstetrics delivery room to meet the

parents when an infant is born with a CL/P, to provide them with support and information on the rehabilitation of the infant. This may be considered a major psychological preventive intervention. However, due to the difficulty of reconciling time information and listening to every family with obstetric or surgical service program, more research is needed in order to design specific and targeted psychological interventions for infants with CL/P and their parents.

In this regard, the current study aimed to describe the infant social withdrawal behavior, the psychological impact on parents, and the parent-infant interaction depending on the different occurring circumstances: type and laterality of CL/P, timing of diagnosis, and timing of the repair surgery. In addition, the present research includes the assessment of the psychological impact of having an infant with CL/P on the fathers, as has been suggested by previous studies (e.g., Weigl, Rudolph, Eysholdt, & Rosanowski, 2005).

It should be mentioned that this study is embedded in a larger French research titled "*Développement relationnel des enfants porteurs de fentes labio-palatines: influence du délai précédant la première intervention chirurgicale et de la perception psychologique de l'anomalie par les parents*", and headed by Dr. Bruno Grollemund, orthodontist, PhD in Philosophy and member of the Faculty of Dentistry in the University of Strasbourg. His experience following children and families with CL/P led him to realize that these children tend to utter very few sounds compared to other children. As a stomatologist treating children having CL/P throughout their development, Dr. Grollemund has observed the psychological difficulties faced by both children and parents, due to the massive threat to narcissism posed by the CL/P, as well as its impact on the intergenerational psychological transmission process (Grollemund et al., 2012a).

This investigation served as a true Action Research project, as it highlighted the great variability of therapeutic protocols throughout regions and countries. More to the point, this project was the first to assess the consequences of the CL/P by simultaneously assessing the social behavior of the infant and the mental health of parents during their evolution and coping with this major traumatic event. The research project also emphasized the need to assess the impact of the malformation on the family life, the psychological distress on parents, maternal and paternal depression, as well as marital satisfaction within the parental couple, in order to provide adequate psychological assistance to everyone in need.

Within this large research project, my assignment consisted of describing the quality of parent-infant relationship, the psychological state of the parents –according to the variables previously mentioned- and of the infant over time, using the concept of social withdrawal behavior as an alarm signal indicating psychological distress and a lasting difficulty in the parent-infant interaction. The impacts of the CL/P on the infant, on both parents, and on the parent-infant relationship were assessed independently.

My contribution to the research project was to evaluate the infant social withdrawal behavior through the ADBB and the m-ADBB scales, and to describe the profiles of the impact on infants, parents and the parent-infant relationship depending on the type and the laterality of CL/P, the timing of the diagnosis, and the timing of the repair surgery. All this with the purpose of finding indicators that allow for the early detection of families at higher risk of mental health problems and that guide psychological prevention and intervention practices.

Accordingly, this thesis consists of six chapters in which is detailed the state of the art, the method, the results, the discussion, the references, and the appendix. The first chapter presents the state of the art

including the main literature about the Cleft Lip and Palate malformation (CL/P), the infant social withdrawal behavior, the quality of parent-infant relationship, and the contributions about the psychological distress on parents having an infant with a congenital malformation, specifically CL/P.

The second chapter shows the method, in which, firstly, it is detailed the objectives and the hypothesis. Secondly, the main descriptive characteristics of the participants are presented. Thirdly, there is a description of each of the instruments of evaluation used in this research, as well as their analysis of internal consistency. Then, is detailed the procedure carried out, the independent and dependent variables, the design and the data analysis.

The third chapter shows the results of the current study. Firstly, we detailed the descriptive outcomes of the analysis of the items, and secondly, the analysis of the total scores for each instrument. Then, it presents the results of the analysis of variance (ANOVA) for repeated measures evaluating the infant social withdrawal behavior, the psychological impact on parents, and the quality of parent-infant relationship, at four and at twelve months postpartum. Also, in this chapter is described the outcomes of the analysis of variance (ANOVA) between groups according the type of the malformation (CLP: Cleft Lip and Palate, or CL: Cleft Lip), the laterality of the malformation (unilateral or bilateral), the timing of diagnosis (prenatal or postnatal), and the waiting time prior to the first repair surgery (early or late).

The fourth chapter presents a discussion about the findings, but also it offers conclusions, some practical implications resulting of this research, as well as the strengths and limitations of the study. Besides, chapter five details the references used and on which this study is based. Finally, in chapter six a glossary which comprises the abbreviations used in the redaction of this thesis is presented in order

to facilitate the interpretation of this study. Additionally, an example of the informed consent used in the recruitment of the sample, as well as examples of the evaluation instruments used in this research.

In sum, this thesis aims to contribute to the understanding of the experience of infants with CL/P and their families, offering a psychological approach of the Cleft Lip and Palate (CL/P) malformation from an evidence-based practice perspective (EBP), and providing data that have a direct implication in clinical practice.

1. STATE OF THE ART

1.1. Cleft Lip and Palate malformation

1.1.1. Definition

Clefts of the lip with or without cleft palate (CL/P) are congenital anomalies immediately recognizable because of the disruptions of normal facial structure. A cleft lip is defined as an abnormal opening in the lip resulting from a failure of the parts of the lip to come together during fetal development (Kummer, 2014). This is, a cleft lip (CL) involves one or more clefts in the upper lip due to a failure in the two sides of the upper lip and nose to fuse correctly (Anderson, 1998).

On the other hand, a cleft palate (CP) involves a fissure in the midline of the roof of the mouth where the two sides of the palate have failed to join normally (Anderson, 1998). Cleft lip and palate can either co-occur or occur independently of each other. Therefore, CL/P has been divided into cleft lip only (CL), cleft palate only (CP) and cleft lip and palate (CLP) (Fraser, 1955). Moreover, the disruption may be unilateral or bilateral, depending if the congenital split is in one (unilateral) or both sides (bilateral) of the upper lip.

Cleft lip and palate (CL/P) results from failures in the frontonasal and maxillary prominence fusions during the fourth and the twelfth week of neonatal development (Corbo Rodríguez & Torres, 2001). While a cleft lip may present serious cosmetic problems, a cleft palate may have more serious consequences principally affecting feeding, hearing, speech and esthetics (Kummer, 2014).

1.1.2. Epidemiology

According to Dixon, Marazita, Beaty and Murray (2011), the common forms of CLP involve disruption of tissue above the lip, extending into the nares and/or the palate. Generally, epidemiologic reports show a trend of 25% of cleft lip (CL), 50% of cases include clefts of the lip with palate (CLP), and 25% cleft palate (CP) (Conway et al., 2015; Corbo-Rodríguez & Torres, 2001). Unilateral clefts are nine times as common as bilateral clefts, and occur twice as frequently on the left than the right. Males are predominantly affected by cleft lip with or without palate (CL/P), whereas females are more commonly affected by cleft palate only (Goodrace & Swan, 2008).

Clefts can either be of non-syndromic origin, where the etiology is unknown, or syndromic, where the cleft is part of a known syndrome. Syndromic CL/P malformations are associated with a range of other congenital malformations and more diverse outcomes. About 70% of cases of CL/P occur as non-syndromic entities, thus, with no other apparent cognitive or craniofacial structural abnormalities or other malformation (Jones, 1988; Marazita et al., 2002).

A cleft, affecting the lip and/or palate, is one of the most frequent birth defects and the most common cause of congenital anomalies in infants (Canfield et al., 2006; Grollemund et al., 2010, 2012a, 2012b; Martelli et al., 2015). The prevalence of CLP is ranging from 1/500 to 1/2500 live births, with differences according to ethnicity, sex, and type of cleft.

In general, Asian and Native American populations have the highest reported birth prevalence rates (1/500), European-derived populations have intermediate prevalence rates at approximately 1 in 1000, and African derived populations have the lowest prevalence rates (1/2500) (Dixon et al., 2011). In France, the prevalence of CL/P is approximately 1 in 700 live births (Grollemund et al., 2012a).

1.1.3. Etiology and risk factors

The etiology of CL/P is multifactorial involving a complex influence of environmental and genetic factors (Jugessur & Murray, 2005). It has been difficult to identify specific etiologic factors because the defects arise early in the embryological development, and because the recurrence rates are modest (Dixon et al., 2011). Whereas twin studies and familial clustering studies have provided compelling evidence for a genetic component to non-syndromic CL/P (Mitchell, 2002), few pedigrees show clear-cut Mendelian inheritance and most cases appear to be sporadic (Jugessur et al., 2009).

Moreover, CL/P is known to be influenced by environmental risk factors (Murray, 2002; Mossey & Little, 2009). In this line, maternal smoking during the first trimester of pregnancy has been repeatedly associated with an increased risk of clefting, and meta-analysis strongly supports an overall odds ratio (OR) for having CL/P of 1.3 among offspring of mothers who smoke (Little, Cardy, & Munger, 2002; Shi et al., 2007; Shi, Wehby, & Murray, 2008). The increased risk resulting from exposure to maternal smoking during the peri-conceptual period raises the possibility that genes in certain metabolic pathways may have a role in the development of CL/P (Dixon et al., 2011).

Additionally, exposure to maternal alcohol consumption has also been suggested as a risk factor, but the evidence has been more inconsistent (Mossey & Little, 2009). Studies suggest that patterns drinking of high doses of alcohol in short periods of time increase risk of clefting (Deroo & Wilcox, 2008). Furthermore, some specific teratogens (Abbott, 2010; Mossey & Little, 2009; Murray, 2002), for example, valproic acid –used to treat epilepsy and bipolar disorder and to prevent migraine headache- have yielded evidence of association with cleft palate (Jentink et al., 2010).

In the same way, nutritional factors, such as folate deficiency, have also been proposed to influence risk of CLP, based on both observational studies and interventional trials using folate supplementation to prevent recurrences of CLP in families (Wehby & Murray, 2010). Likewise, food fortification programs using folic acid have revealed detectable decreases in the rates of clefting in some (Yazdy, Honein, & Xing, 2007; Johnson & Little, 2008) but not all studies (Ray, Vermeulen, Wyatt, & Cole, 2003; López-Camelo, Castilla, & Orioli, 2010). Furthermore, some data suggested roles for zinc deficiency in risk of oral clefts in populations in which zinc status is highly compromised (Munger et al., 2009), for cholesterol deficiency in facial clefting (Porter, 2006), and for multivitamins in general in cleft prevention (Johnson & Little, 2008).

Other environmental exposures have been evaluated for possible roles in clefting, including hyperthermia (Shahrukh, Gallaway, Waller, Langlois, & Hecht, 2010), stress, maternal obesity, occupational exposures, ionizing radiation and infection (Mossey, Little, Munger, Dixon, & Shaw, 2009). Nevertheless, there is no consensus yet on the harmful effects of all these factors, and prospective studies large enough to measure effects on a relatively rare disorder such as clefting may be required (Dixon et al., 2011). Finally, it should be noted that pregnancy planning has been shown to have a protective effect (Mossey, Davies, & Little, 2007).

1.1.4. Diagnosis

Since the 1980's, prenatal detection of CL/P has been possible with a transabdominal ultrasound (Maarse et al., 2010). Nowadays, the diagnosis of CL/P malformation is more common during the prenatal period because imaging techniques have improved in 3D and 4D. However, diagnosis of CL/P can still be revealed at birth or soon after birth (Grollemund et al., 2012a; Habersaat et al., 2013).

In 1981, Christ and Meininger reported the first case of cleft lip and palate diagnosed with antenatal ultrasound. Currently, prenatal diagnosis of cleft lip by ultrasound can be performed as early as 12 weeks into pregnancy, and the infant's prognosis is generally considered to be dependent upon the presence and type of associated anomalies (Rey-Bellet & Hohlfeld, 2004). When the cleft is discovered, the ultra-sonographer turns to the multidisciplinary cleft team for counsel and care of the expectant parents, and for planning the reparative surgery and dental care.

The announcement of a prenatal or postnatal diagnosis of a congenital anomaly suddenly and unexpectedly disrupts the parent's expectations of a healthy child (Lawoko & Soares, 2006). It has been suggested that prenatal diagnosis may constitute a long-lasting psychological stressor for parents given the increased latency period between diagnosis and treatment availability (Brosig et al., 2007).

Sharp, Strauss and Lorch (1992) found that, even though the parents are shocked when they learn that their future child has a cleft, the prenatal detection is often seen as advantageous. It makes it possible for the parents to go through the grieving process and to deal with distressing emotions during pregnancy rather than during the more chaotic time around the birth of their child (Rey-Bellet & Hohlfeld, 2004; Davalbhakta & Hall, 2000). Besides, it has been shown that the

prenatal diagnosis has a positive impact on the future mother-child relationship (Baker, Owens, Stern, & Willmot, 2009).

Similarly, Rey-Bellet and Hohlfeld (2004) observed, in a retrospective study, that 96% of parents considered prenatal diagnosis to be beneficial. In this sense, Davalbhakta and Hall (2000) suggest that a prenatal diagnosis -supported by antenatal counselling- improves the psychosocial aspect of cleft care and can make the delivery of a baby with a cleft a positive experience.

Moreover, a prenatal diagnosis also benefits the medical team that corrects the cleft lip, since anticipating the birth of a baby with CLP helps in provisionally listing the infant for lip repair (Davalbhakta & Hall, 2000). Although some studies have found that most parents prefer to know about the diagnosis during the prenatal period (Kuttenberger, Ohmer, & Polska, 2010; Rey-Bellet & Hohlfeld, 2004), other families consider the knowledge of the cleft to ruin the pregnancy (Rey-Bellet & Hohlfeld, 2004).

In this regard, several mothers said that they would have preferred to know the diagnosis later in pregnancy, at 7th or 8th month of pregnancy, as they would still have enough time to recover from the initial shock. This timing of diagnosis was short enough to keep their anxiety under control and not ruin the pregnancy (Maes, Demey, & Appelboom-Fondu, 1998).

As can be seen, studies are not conclusive about this issue, and screening of fathers is uncommon. In this line, Zeytinoglu, Davey, Crerarnd and Fisher (2016a) insisted on the importance of studying the impact of the prenatal and postnatal diagnosis on parents because distress can affect how they attach to their infants, and it has been found that poor attachment is potentially detrimental to later psychological functioning among children born with CLP (Fox,

Nordquist, Billen, & Savoca, 2015; Pope, Tillman, & Snyder, 2005; Speltz, Armsden, & Clarren, 1990).

A better understanding of how the timing of diagnosis affects both mothers and fathers could lead to the development of family centered approaches to pediatric care (Zvara, Schoppe-Sullivan, & Dush, 2013).

1.1.5. Implications on infants

Even though cleft lip and palate malformation is not a major cause of mortality in developed countries, it causes considerable morbidity to affected children and imposes a substantial burden on the health, quality of life and socio-economic well-being of children and their families (Wehby & Cassell, 2010).

a) Functional problems

Individuals with cleft lip and palate may experience functional problems with feeding (swallowing and chewing), speaking (phonation), hearing and ventilation. Early in life, affected individuals are at an increased risk of experiencing difficulties related to feeding (Nackashi, Dedlow, & Wood-Dixon, 2002).

Feeding the infant with CLP is one of the first care challenges faced by the parents and caregivers. Because of the opening in the palate, the infant is unable to create intra-oral pressure required to suck effectively. This may result in inadequate nutrition, lengthy feeding time, nasal regurgitation, choking, coughing and excessive air intake. Frequently, therapeutic intervention is required to assist the parents to feed the infant so as to encourage adequate weight gain (Kummer, 2014).

Moreover, it has been demonstrated that communication problems related to CL/P are noticeable from a young age. Velopharyngeal dysfunction, frequently associated with cleft palate may result in a

variety of speech disorders including impaired speech articulation, nasal emission of air, nasal resonance and voice disorders (Peterson-Falzone et al., 2010).

For example, Neiman and Savage (1997) found that toddlers with CL/P showed a delayed development in the expressive language domain at 36 months. Similarly, children with CL/P have an increased risk for dyslexia related to fluency and naming speed (Yazdy, Honein, Rasmussen, & Frias, 2007).

When the malformation extends to the palate, the sound of the voice can be modified, nasal or hoarse. Additionally, it has been shown that speech difficulties are due to a very short palate, which retards the flow and the speed of speech on these children (Richman & Ryan, 2003). All of these problems may result in defective speech, language and hearing skills, and may have a negative impact on communication and educational progress (Peterson-Falzone et al., 2010).

Furthermore, it was found that velar insufficiency, the existence of residual palatal or buccal vestibular holes, lead to false nasal food routes causing chronic rhinitis. Besides, other morphological disturbances -deviation of the nasal septum, disturbance of the nasal valve, and the presence of an ectopic tooth in the floor of the nasal cavity- compromise nasal ventilation and tubal function. Consequently, the incidence of acute otitis media is recurrent, complicated by a deafness of transmission that is more or less severe and very often bilateral and lasting (Grollemund et al., 2010).

It has been found that these functional problems could result in poorer cognitive functioning (Roberts, Mathias, & Wheaton, 2012), in a delay in reading acquisition for nearly 30-40% of children with CLP (Richman, Wilgenbusch, & Hall, 2005). Likewise, academic difficulties are often seen with these children, as approximately 25% of CLP children repeat

grades or leave the school prematurely (Broder, Richman & Matheson, 1998).

b) Psychological issues

Several studies have reported psychological issues among infants, children, adolescents and young adults with CLP, for example in self-perception, social skills, overall adjustment, social inhibition (Kapp-Simon et al., 1992; Kapp-Simon & McGuire, 1997), interactional competencies (Brand et al., 2009), behavioral problems, satisfaction with facial appearance, depression, and anxiety (Hunt, Burden, Hepper, & Johnston, 2005).

Different physiological and sociocultural factors contribute to the development of psychosocial concerns among individuals with any form of facial anomaly (De Sousa, Devare, & Ghanshani, 2009). Self-perception, which plays a fundamental role in influencing an individual's self-esteem and psychological adjustment, can be affected by cleft lip and palate (Strauss, Broder, & Helms, 1988). Additionally, parental influence also shapes ones psychosocial perception.

The attitudes, expectations and degree of support shown by parents can influence a child's perception of his/her cleft impairment (Bull & Rumsey, 1988; Lansdown, Lloyd, & Hunter, 1991). Generally, parents of children with clefts may be more tolerant of their child misbehavior and are more likely to spoil their child by being overprotective (Turner, Thomas, Dowell, Rumsey, & Sandy, 1997; Harper & Richman, 1978). This could contribute to behavior problems in childhood.

Furthermore, speech and language disorders, hearing loss and concerns about esthetics are thought to contribute to these psychological challenges (Hunt et al., 2005; Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Patrick et al., 2007; Thomas, Turner, Rumsey, Dowell, & Sandy, 1997). Many children with cleft lip and

palate may have a less attractive facial appearance or speech than their peers. Kummer (2014) suggested that as a result of poor physical appearance and poor communication skills, the child may experience teasing and bullying, leading to poor self-perception, and may find it difficult to make friends and to progress academically in school.

A face that has been operated can remain marked and scarred. There is often an asymmetry of the upper lip and nose, and, for the most severe forms, an obvious disproportion can be seen between the different stages of the face seen in profile (Grollemund et al., 2010). In this regard, Hunt, Burden, Hepper and Johnston (2005) found a high incidence of teasing over facial appearance among children with cleft lip and palate.

Likewise, Topolski, Edwards and Patrick (2005) observed that children with visible facial differences have lower quality of life, compared with children with no chronic conditions. Similarly, Wheby, Ohsfeldt and Murray (2006) found a modest decrease in the health-related quality of life among children with CL/P. In this line, Murray et al. (2010) observed that children with clefts showed high rates of teacher-reported social problems, and anxious and withdrawn-depressed behavior. Direct observations also revealed difficulties in social relationships. Interestingly, these effects were explained by communication problems.

Regarding long-term outcomes, adolescents with CL/P showed social inhibition (Kapp-Simon & McGuire, 1997), and those with low social competence reported loneliness and social anxiety (Pope & Ward, 1997). In addition, it has been reported that individuals affected with CL/P have lower income, lower marriages rates and older age at marriage (Ramstad, Ottem, & Shaw, 1995).

Besides, increased social anxiety among affected adults has also been found, however these findings varied between studies (Berk, Cooper,

Liu, & Marazita, 2001; Cheung, Loh & Ho, 2007). It has been found a higher risk of hospital admission due to mental health complications among adults with CL/P (Christensen & Mortensen, 2002).

Furthermore, a higher risk of cancer, cardiovascular events, and a significantly increased mortality rate due to suicide among patients with CLP was reported compared to unaffected individuals (Christensen, Juel, Herskind, & Murray, 2004). Additionally, alterations in child bearing patterns have been reported among women with clefts (Yttri, Christensen, Knudsen, & Bille, 2011). However, cleft lip and palate malformation affects not just the infants, but also parents and family (Zeytinoglu & Davey, 2012).

1.1.6. The several therapeutic interventions: a long and complex journey awaits each family having an infant with CL/P

Treatment for cleft lip and palate is typically characterized by extensive examination aimed at identifying: 1) physical anomalies, 2) feeding and swallowing disorders, 3) hearing disorders, 4) genetic and other associated disorders, and 5) the need for psychological support for the patient and his/her family (Kasten et al., 2008). Consequently, children are treated by multidisciplinary teams of specialists at birth and continuing through early adulthood (American Cleft Palate Craniofacial Association, 2009).

Interventions and treatments for infants with CL/P can vary depending on the severity of the cleft, the presence of associated syndromes and/or other birth defects, and the infant's age and needs (Nackashi, Dedlow, & Wood-Dixon, 2002). For example, children with a cleft lip can breast-feed, they need little specific care and few surgeries. On the other hand, a bilateral cleft lip and palate requires many episodes of surgery, is accompanied by feeding difficulties (e.g., need of a removable palate appliance to avoid nasal reflux of milk, impossibility

of breast-feeding efficiently), and has a greater impact on facial appearance (Chetpakdeechit, Hallberg, Hagberg, & Mohlin, 2009).

CL/P patients require different types of interventions, including dental and stomatological treatment, speech therapy, psychosocial intervention and numerous repair surgeries depending on the seriousness of the malformation. The first intervention is on the lip, and this is the only one surgery performed during the first year of life.

Consistent with Dissaux et al. (2015), primary cleft surgery must ensure that optimal aesthetic and functional (speech development) results are obtained, with minimal negative effects on facial growth. It is important to consider that cleft treatment is multidisciplinary, but the surgeon, acting first, sets the tone over the whole of an infant's life until adulthood.

According to Guedeney, Le Foll, Vannier, Viaux-Savelon and Wendland (2014), the pioneer surgeon on CL/P repair intervention was Victor Veau (1871-1949). It was from a case of re-education of a girl with cleft lip and palate, led by Susanne Borel-Maisonny. Speech therapy emerged in France as a result.

Currently, the modalities of reconstructive surgery vary according to the severity of the malformation and the protocol followed by each surgical team. Nowadays, no generic protocol has been recognized by the entire medical community, therefore, each patient is cared for according to the experience and the decisions of the medical team in charge.

In Europe there are more than two-hundred reference centers for handling children with CL/P. This yields the use of 190 different surgical protocols. However, there is no protocol that proved to be superior to the others (Dissaux et al., 2015; Grollemund et al., 2012a; Shaw et al., 2001). These discrepancies can be explained by the fact that the

aesthetic and functional outcome of a protocol can only really be evaluated in adulthood, when the child's growth is complete.

In France, the planning of the surgical intervention varies by medical center. Certain teams prefer early intervention, immediately after birth, in order to quickly restore functions (ventilation, swallowing and phonation) and to reduce the aesthetical impact of the defect on the parents and the family. In this line, Galinier et al. (2008) suggested that early intervention to close the cleft lip is essential for the psychological well-being of the parents.

Other medical teams prefer to wait three of even six months to perform the lip repair surgery. By delaying the intervention, these teams aim to make use of the particularly fast physical growth during this period. The individualization of the different part of a muscle is thus easier, and this improves the precision and the quality of the surgical gesture (Grollemund et al., 2012a). Nevertheless, Goodacre, Hentges, Moss, Short and Murray (2004) found that surgical outcome appears unaffected by this difference in intervention timing. Besides, related to the higher risk of anesthesia in the neonatal period, Galinier et al. (2008) did not found any literature that reported an anesthesia risk that was greater in the neonatal period than at three months in patients without risk of complications.

However, the psychological consequences of early vs. later lip repair may differ, and were largely unknown before this research was planned. In this regard, there is also an urgent need to assess the timing of interventions and surgeries to determine if earlier interventions improve infant’s outcomes (Yazdy, Honein, Rasmussen, & Frias, 2007), and reduce the impact on the family, which is clearly a major issue.

1.2. Social withdrawal behavior in infants

1.2.1. Introduction

Developmental psychology has taught us that infants are social beings, actively seeking interactions and contact, both physically and psychologically, with their environment (Fogel, 1993). Infants are born with social and cognitive capacities that enable them to participate in human interaction (Stern, 1985; Trevarthen & Aitken, 2001). However, it is important to consider that infant social behavior develops in the context of early parent-infant interaction (e.g., Feldman, 2007).

Infant's interaction skills include the ability to initiate and maintain eye contact with another person, to vocalize, to use and to imitate facial expressions, and to use body and head movements to initiate and maintain an interaction or provoke a reaction in others (Field, Cohen, Garcia, & Greenberg, 1984; Meltzoff & Moore, 1977; Trevarthen & Aitken, 2001).

This is to say, right from birth, infants possess abilities for initiating, maintaining and terminating interactions. They are motivated to interact with the caregiver and they actively seek social stimulation during periods of awareness (Trevarthen & Aitken, 2001). Observational studies of young infants have suggested that their behavior and facial expressions, even at early ages, are units of expression with a level of organization that are embedded with information about children's internal states and intentions, which allow them to communicate their needs to adults (Tronick, Als, & Brazelton, 1980; Weinberg & Tronick, 1994).

Therefore, facial expression and vocalizations are pre-verbal efforts of the infant to communicate, and they are indicators of feeling states. Most infants show three emotions by 2 months of age (interest, contentment, and distress) and eight emotions by 7 months (joy,

contentment, anger, disgust, surprise, interest, distress, and sadness) (Izard, Huebner, Risser, McGinness, & Dougherty, 1980). Smiling in response to a person's voice and face occurs by 8 weeks old, while anxiety towards strangers is generally considered to be evident by around 9 months of age (Snow, 1998).

Similarly the ability to respond to stimulation offered by others is an essential part of early non-verbal communication, as it acknowledges the presence of the other person, and communicates whether one is available or not for interaction. These signs are part of the opening and closing of circles of communication (Greenspan, 1992). It is important to consider that, in typical social development, even though infants may differ in their style and degree of responsiveness to various stimuli (i.e., have different temperaments), they are still responsive to social interaction with an adult (Fox, 2004).

Moreover, eye contact is an important sign of normal development in infants (Farroni, Csibra, Simion, & Johnson, 2002), and generally it is the first feature that is affected by any adversity the infant may experience. Eye contact is also the main mode of establishing an early relationship between a parent and an infant. All these social skills are signs for the evaluation of social withdrawal, considered by Greenspan (2000) as a risk factor for infant development.

1.2.2. Definition

Social withdrawal behavior is defined as "a chronic diminution of the attachment system, which is gradually generalized into a diminished engagement, and lowered reactivity to the environment at large" (Dollberg, Feldman, Keren, & Guedeney, 2006; p. 295). Social withdrawal behavior, from birth onwards but clearly as early as 2 months of age, is indicated by diminished or lacking of either positive (e.g., smiling, eye contact, cooing) or negative behaviors (e.g., dampening of protest, and diminished crying) (Guedeney, 1997).

According to this author, sustained social withdrawal is similar to a state of learned helplessness.

Dollberg et al., (2006) argued that sustained withdrawal behavior has its roots in the organism's evolutionary, biological-temperamental repertoire, as evidenced from animal studies. They suggested that under certain contextual circumstances, the natural mechanism of episodes of social withdrawal may lead to the development of a defensive strategy of sustained withdrawal, which acts against the infant's natural tendency to reach out to the social environment.

Microanalytic studies have shown that brief episodes of infant withdrawal appear frequently during parents–infant interactions, playing an important role in the regulation of early interactions (Beebe, Lachmann, & Jaffé, 1997; Brazelton & Cramer, 1990; Greenspan & Wieder, 1993; Weinberg & Tronick, 1994). Closing the eyes, turning the head and looking away are the infant's means of regulating the flow of interaction (Tronick, 1989).

This is, infants might subsequently develop sustained social withdrawal behaviors as a way of handling repetitive or durable violations of his or her expectations within social interactions (Murray & Trevarthen, 1985; Puura et al., 2010). If the interaction is continually too intrusive or unresponsive, infants may learn that they have to consistently withdraw to reduce their arousal level (Tronick & Weinberg, 1997). Over a longer period of time, stronger protesting behavior can lead to increasingly pathological behavior in terms of avoidance, as observed in the Still Face experiment (Cohn & Tronick, 1987).

1.2.3. History of the concept

René Spitz (1946) was one of the first authors to use this term in his clinical description of anaclitic depression in young infants aged around nine months. Then, in 1952, Robertson and Bowlby described a three stage emotional reaction in young children —protest, despair, and withdrawal behavior—and eventually detachment in face of prolonged separation.

Subsequently, Engel and Reichsman (1956, 1979) described sustained social withdrawal behavior as a defense mechanism in a 14 months infant, during a long hospitalization. Based on this case, Engel and Schmale (1972) developed the concept of conservation-withdrawal threshold as a biological mechanism which allows the system, under certain severe conditions, to disengage in face of the external environment in the service of conserving energy and assuring the organism's survival.

Meanwhile, Tizard (1977) and Tizard and Rees (1975) observed that institutionalized children who had been admitted to a residential nursery before the age of 4 months were largely unresponsive and emotionally withdrawn at 4 years. Then, in 1982, Fraiberg described a group of pathological defenses -avoidance, freezing and fighting- observed in infants between 3 and 18 months of age who experienced severe danger and deprivation. These early defenses are embedded in the biological repertoire, and they are related to the concept of withdrawal.

Moreover, another study that has contributed to the development of social withdrawal as a construct, is the description of two infants with non-organic failure to thrive (NOFTT). This study highlights the possibility to a better understanding of NOFTT within the context of conservation-withdrawal. Infants with failure to thrive (FTT) are described as sad, depressed, withdrawn and irritable (Powell & Low,

1983). Indeed, findings confirm that some FTT children prefer interacting with inanimate objects (Rosenn, Loeb, & Bates, 1980), make little eye contact, rarely vocalize, seem to dislike cuddling, and may engage in self-stimulatory activities, or in rumination (Powell & Low, 1983). Currently, all these are signs of social withdrawal behavior.

More recently, Panksepp (2006) proposed a schema of the main types of emotional systems in mammalians: lust, care, panic, play, fear, rage, and seeking. In this proposal, withdrawal behavior has been conceptualized as part of the panic and fear systems. In addition, Rubin, Coplan and Bowker (2009) proposed that the ontogeny of a socially withdrawn profile begins with newborns who are biologically predisposed to have a low threshold for arousal when confronted with social (or nonsocial) stimulation and novelty.

Furthermore, Costa and Figueiredo (2012) identified three psychophysiological profiles in infants (withdrawn, extroverted and under-aroused) using the ADBB scale, the NBAS scale, and cortisol levels in saliva before and after a vaccine. The withdrawn profile (17% of the sample) was identified with a below-average neurobehavioral organization, with signs of social withdrawal, and above-average endocrine reactivity. The extrovert profile (57% of the sample) was characterized by an above-average neurobehavioral organization, with no signs of withdrawal, and slightly above-average endocrine reactivity. On the contrary, the under-aroused profile (26% of the sample) was identified as having a neurobehavioral organization similar to the mean, certain signs of withdrawal, and below-average endocrine reactivity.

In the withdrawn profile, a worse performance was observed in the evaluation of neurodevelopment compared to the other two profiles. With regard to general interaction, withdrawn babies were more difficult, serious and unsatisfied, and less involved with the

environment. In addition, the mothers of the withdrawn babies presented more depressive behavior in the interaction, were less attentive and communicative and their interaction was of lower quality. All the literature described above has contributed to the development of the construct of social withdrawal in infants.

1.2.4. Prevalence

Studies indicated that the prevalence of infant social withdrawal, evaluated thought the ADBB scale, ranged from 39.4% in infants from immigrant families with low socio-economic status (Burtchen et al., 2013) to 7% in general population (Puura et al., 2010).

In France, Guedeney, Foucault, Bourgen, Larroque, and Mentré, (2008) found 13% of infants with sustained social withdrawal in a sample of 640 infants free of charge with a mean age of 16 months. In addition, in a longitudinal study it was observed a 14% of socially withdrawn infants (Guedeney et al., 2012). Moreover, in a recent study conducted by Tauber et al., (2017) in infants with a genetic diagnosis of Prader-Willi Syndrome, the prevalence was of 65% of social withdrawal. These finding leads to the necessity of identify the factor risk for social withdrawal.

1.2.5. Risk Factors

It has been found that social withdrawal occurs more frequently in adopted male babies, in infants born of twin pregnancy or living in risk conditions -in shared custody or in foster care- (Guedeney et al., 2008). Moreover, Espié et al. (2011) found in a sample of institutionalized infants that 33.8% had signs of social withdrawal at the time of admission to the institution and that this percentage remained stable during follow-up at two, four and twelve months, except for the nine months, when this percentage increased almost to 50% of the babies. In addition, in this same study, it was observed

that among babies with delayed psychomotor development, 81.3% showed signs of social withdrawal.

Similarly, maternal psychopathology, difficulties in parent-infant interaction, and a stressful family life situation, were found to be recurrent factors in cases of infant social withdrawal in a study conducted by Puura et al. (2010). Moreover, Molteno, Jacobson, Carter, Dodge and Jacobson (2014) found that alcohol consumption during pregnancy was associated with greater social withdrawal in infants and with a decrease in infant's activity.

Other risk factors that have been found to be associated to social withdrawal are prematurity (Braarud et al. 2013; Moe et al., 2016), sleep and feeding disorders (Dollberg et al., 2006), somatic suffering (Puura et al., 2010), mother's current depressive symptoms and father's perceived moderate or poor mental health (Mäntymaa et al., 2008; Puura et al., 2013); maternal symptoms of irritability, anxiety, and sadness (Matthey et al., 2005), maternal psychopathology (De Rosa et al., 2010).

Risk factors related to parent-infant interaction have been also identified. For instance, Dollberg et al. (2006) observed more negative relational patterns in mother-withdrawn infant dyads in terms of higher maternal intrusiveness, lower reciprocity, and lower infant involvement. These poor mother-infant interaction patterns, when infant social withdrawal is present, were also found in other studies (e.g., Costa & Figueiredo, 2013; De Rosa et al., 2010; Puura et al., 2007; Rochet & Mellier, 2007).

Concerning infants with clefts, it has been found that these infants spend less time vocalizing, smiling and looking at their mothers, as compared with healthy infants (Speltz, Goodell, Endriga, & Clarren, 1994). More recently, these findings were confirmed and extended by Montirosso et al. (2012) and Murray et al. (2008) noticing that infants

with cleft lip showed fewer communicative signals toward their mothers, producing fewer positive vocalizations, spending less time in visual contact with their mothers, engaging less in active exploring of the environment and exhibiting more self-absorbed behavior than controls (Montirosso et al., 2012; Murray et al., 2008). These outcomes lead to our interest in evaluating social withdrawal in infants born with cleft lip and palate (CL/P).

Moreover, it is important to consider that infant social withdrawal, by definition, makes the infant less available for interaction, either because of factors relating to their own capacity to regulate stimulation or factors relating to the care, such as experiencing their caregiver as emotional unavailability (Solomon & George, 1999). Moreover, social withdrawal leaves infants unavailable for the developmental opportunities afforded by the interpersonal space. Therefore, it is necessary to know the implications of social withdrawal behavior on infant development.

1.2.6. Implications of social withdrawal for infant development

Studies found associations between social withdrawal, in the first months of life, and difficulties in the later development. One of the most relevant studies indicate a relationship between social withdrawal -at six months of age- and deficits in social and communication skills (assessed by the Behavior Assessment System-2), as well as cognitive and language skills (assessed with Bayley-III Scale) at 30 months of age (Milne, Greenway, Guedeney, & Larroque, 2009). In the same study, positive associations were also found between social withdrawal -at six months of age- and the tendency to unusual or peculiar behaviors, such as disconnection or lack of interest in the environment, as well as attention problems (evaluated with the Behavior Assessment System-2) at two years of age.

In addition, it has been observed that social withdrawal -at 12 months of age- corresponded to relational problems at 3 years and to behavioral alterations at 5 years (evaluated with the Strength and Difficulty Questionnaire) (Guedeney et al., 2014). Similarly, the acquisition of motor and language skills was lower in children with social withdrawal at 12 months (Guedeney et al., 2016).

Furthermore, it has also been shown that children diagnosed with Fetal Alcohol Syndrome -at five years- showed greater social withdrawal and lower level of response and activity at six months of age (Molteno et al., 2014). Besides, these authors observed that social withdrawal was the strongest predictor of children's IQ at nine years. All these findings evidence the negative effects of infant social withdrawal on subsequent development.

In sum, development is an active process, and the optimal mental development within the infant's potentialities is not achieved when the infant is withdrawn for a period of time. Thus, social withdrawal behavior in infants is an important alarm signal to screen for, in the context of preventive services and of clinical treatment settings (Glascoe & Macias, 2003; Guedeney et al., 2011).

Early identification of infants at risk allows for early inquiry about its cause, either in infants, parents or interaction factors. Accordingly, comprehensive assessment and treatment approaches for families need to include screening of parents during infants evaluations and to explore parent–infant relationship (Vidair et al., 2011). One of the most important tasks in the field of infant psychopathology is to identify the relationship disturbances between parents and their infants.

1.3. Parent-infant relationship

The early parent-infant interaction provides the scaffolding needed by the infant to develop (Brazelton, Koslowski, & Main, 1974; Fox, 2004). The relationship established between the infant and his/her parents is the earliest and closest among the many relationships that individuals experience throughout their life. These interactions are fundamental to the lives of both parents and infants, and provide one of the most important environments in which infants develop as individuals and as members of their culture (Russell, Mize, & Bissaker, 2002). Early parent-infant interaction plays a fundamental role in infant's cognitive and socio-emotional functioning (e.g., Hentges et al., 2011; Murray et al., 2008; 2010).

Transactional theories proposed that the quality of early parent–infant relationships is influenced by characteristics of both the parent and infant (Sameroff, 1993). Additionally, these theories recognize that infants and infant–parent relationships are imbedded in the family context and develop in a reciprocal, bidirectional manner over time (Sameroff & Fiese, 1990, 2000). This is, caregiver's behavior is guided by infant's expressive displays (e. g. facial expressions, gaze, gestures and vocalizations). In turn, infant behavioral and affective states are affected by the expressive displays of the caregiver (Montirosso et al., 2010).

Similarly, the mutual regulation model (MRM; Gianino & Tronick, 1988) argues that the interaction is organized by bidirectional exchange of communicative signals that infant and parents use to coordinate the interaction and to cope with the stress of inevitable interactive lack of coordination. From this viewpoint, the quality of the interaction is influenced by the ability of each participant to cope with external stressors, regulate his emotional states, express communicative

messages, and respond to his partner's affective communications and needs (Montirosso et al., 2010).

Moreover, the quality of early parent-infant interaction has an important influence on the infant's physical, psychological and social development (Sameroff & Fiese, 2000; Stern, 1985). It has been shown that whereas parent and infant interactions characterized by mutual engagement and affective reciprocity enhance the infant's development, maladaptive interaction between parent and infant places the infant at risk for later emotional, cognitive, and behavioral difficulties (Bakeman & Brown, 1980; Field, 1987).

Previous studies have demonstrated that infant–parent interaction sequences typically involve an identifiable beginning, middle, and end (Cohn & Tronick, 1989; Field, 1987). Healthy interactions are characterized by parent and infant establishing joint attention, modulating, or matching voice and facial affect (Nicely, Tamis-LeMonda, & Grolnick, 1999), and gradually decreasing activity to end the interaction.

On the contrary, maladaptive interactions also involve initiation, maintenance, and termination phases, but the interaction's quality is extremely different. Maladaptive interaction is characterized by disengaged or intrusive parental stimulation to which the infant responds with flat or negative affect (Field, 1983; Stern, 1985; Tronick & Gianino, 1986). Parent and infant appear unresponsive to each other.

Over time, such interactions may increase the infant's risk for developing relationship disturbances (Sameroff & Emde, 1989). Similarly, some theorists have suggested that, over time, impairment in reciprocal interaction may evolve from temporary disturbances to more serious relationship disorders, impeding the infant's ability to achieve developmental milestones (Anders, 1989; Emde & Sameroff, 1989).

In this regard, parent-infant relationship has an important role in the process of infant's emotional regulation. For instance, affective communication between the infant and the parents, sharing of affects, and the caregiver's empathetic understanding of the infant's affective state may be seen as prerequisites in the process of emotional regulation (Trevarthen & Aitken, 2001).

In this dyadic process, by avoidance and withdrawal the infant can signal to the caregiver that he/she has had enough stimulation. A sensitive caregiver notices the infant's signals and lowers stimulation's level, so that a balanced state in emotional regulation may be achieved. In this line, Schore (2001) explained that the role of the primary caregiver is to regulate the infant's maturing limbic system, and in this way, attachment relationship facilitates the expansion of the infant's coping capacities.

Furthermore, the ability within the parent–infant triad to synchronize with each another, particularly during the first 18 months of the infant's life, is a key element in early development (Feldman, 2007; Mäntymaa, 2006). According to Feldman (2007, p. 330), synchrony is described as "a time-bound, co-regulatory lived experience within attachment relationships that provides the foundation for the child's later capacity for intimacy, symbol use, empathy, and the ability to read the intentions of others". Likewise, both maternal and infant factors contribute to synchrony between parent-infant interactions.

In other words, synchrony in the context of parent–infant interactions addresses the matching of behavior, affective states, and biological rhythms between parent and child that together form a single relational unit. Feldman (2007) also describes synchrony as the complex 'dance' that occurs during short, intense, playful interactions; builds on familiarity with the partner's behavioral repertoire and interaction rhythms (Beebe, 1982; Fogel, 1993; Stern, 1977; Tronick, 1989). This

is, synchrony is conceptualized in terms of the parent–infant dyad as a temporal and organizing feature of the relationship (Feldman, 2007).

The maternal repertoire during the postpartum period in humans includes gaze at the infant's face, high-pitched vocalizations, affectionate touch, and careful adaptation to the infant's state and signals (Feldman & Eidelman, 2003; Minde, Perrotta, & Marton, 1985). Thus, parents are sensitized and attuned to meet the needs of their newborn (e.g., Brazelton & Cramer, 1990), however psychological distress or mental illnesses are some factors that have been shown to impair the parent's ability to engage and to interact with their infant in a positive way (e.g., Murray, Fiori-Cowley, Hooper, & Cooper, 1996).

Accordingly, infant's characteristics and medical conditions also may contribute to complicate parent-infant relationship. For example, in babies having cleft lip and palate (CL/P), the disfigurement makes infants less appealing (Goodacre, Henteges, Moss, Short, & Murray, 2004) and less easy for parents to interpret their expressions (Field, 1977; Field & Vega-Lahr, 1984; Goldberg, Brachfeld, & DiVitto, 1980). Additionally, in this particular case, parents may experience an initial emotional reaction of shock, anger, sadness and guilt -faced the cleft lip and palate infant's malformation- (Bradbury & Hewison, 1994) that may impair their parental skills.

1.3.1. Parent-infant relationship within the stressing situation of infant cleft lip and palate (CL/P)

At birth, the relationship of any parent with her or his newborn is based on conscious and unconscious emotions which for the most part are transmitted by touch, tone of the voice, gazes and facial expressions. The malformation makes infants with clefts less attractive (Goodacre, Hentges, Moss, Short, & Murray, 2004) and parents have more difficulties interpreting their expressions (Field & Vega-Lahr, 1984), which is a requirement for a healthy mother-infant relationship.

Besides, the malformation can prevent the recognition of an intergenerational affiliation, and hamper the integration of the infant into the family (Strauss, 2002). Several studies have indicated that the interaction style between parents and infants with a cleft lip (with or without cleft palate) (CL/P) differs significantly compared with control dyads (Barden, Ford, Jensen, Rogers-Salyer, & Salyer, 1989; Field & Vega-Lahr, 1984; Koomen & Hoeksma, 1992; Langlois, Ritter, Casey, & Sawin, 1995; Montirosso et al., 2012; Speltz, Goodell, Endriga, & Clarren, 1994; Wasserman, Allen, & Solomon, 1985; Wasserman, Allen, & Linares, 1988).

For instance, Viaux-Savelon, Rosenblum, Mazet, Dommergues and Cohen (2007) found that mothers receiving the announcement of an anomaly during the prenatal period, described a suspension of the infant's investment, with a perturbation of the fetus's representation. In this regard, it is interesting to evaluate if there are differences in the quality of parent-infant relationship according to the timing of the diagnosis (prenatal or postnatal).

Furthermore, it has been observed that mothers of infants with cleft lip were less responsive and sensitive in the interpretation of their infants' signals (Koomen & Hoeksma, 1992; Montirosso et al., 2012). In a play situation, mother-infant interactions were less active, responsive, and playful, as compared with those of control dyads (Field & Vega-Lahr, 1984; Speltz, Goodell, Endriga, & Clarren, 1994; Wasserman, Allen, & Solomon, 1985; Wasserman, Allen, & Linares, 1988).

In turn, children born with a cleft spend less time vocalizing, smiling and looking at their mothers, in comparison to healthy children (Spetlz et al., 1994). Similarly, Habersaat et al. (2013) observed that infants with a cleft were more difficult and less cooperative during interaction at 2 months of age with their mother, compared with infants without a cleft. However, they did not find differences in dyadic interactive styles.

Besides, Montirosso et al. (2012) observed that infants with a cleft lip were less engaged and their mothers showed more difficulty in interaction than control group. This is, the presence of infant clef lip may complicate the mother's interpretation of infant signals, and, in turn, this behavior might lead to less infant involvement in the interaction and more infant withdrawal. Besides, mothers of infants with cleft lip displayed more negative affectivity.

Likewise, Despars et al., (2011) found that mothers of infants with a cleft lip (with or without palate) more often experienced insecure parental working internal models of the child, this is, a fewer balanced and more disengaged attachment representations. Parental representations may affect the dyadic functioning and early parent-infant interactions.

It is important to consider that the type of the malformation will impact differently on parent-infant relationship. For example, in mother-infant face-to-face interactions, Endriga and Speltz (1997) found that mothers of less-impaired infants (cleft palate only) were more distant than mothers of more-impaired infants (cleft lip and palate). These authors proposed that this results could be explained because first mothers received less professional attention and support than those of infants with cleft lip and palate.

On the contrary, Despars et al. (2011) observed that the severity or complexity of the cleft was not related to parental representations and posttraumatic stress. These inconclusive findings lead us to the interest to evaluate the quality of parent-infant relationship according to the type and laterality of the CL/P malformation.

In addition, one factor that may affect parent-infant interactions in the context of infant cleft lip is the timing of surgical repair, since this affects dramatic cosmetic change (Slade, Emerson, & Freedlander, 1999). On the one hand, late repair may be advantageous because

parents have time to adjust to the infant's condition and plan for surgery (Munro, 1995), and parent–infant contacts immediately after the birth are not disrupted by the infant's removal for surgery and post-operative recovery.

Furthermore, family and friend's reactions often made parents wish the surgical repair could be done at the earliest. In favor of early repair is the possible benefit in terms of parent–infant interactions in the initial months (Bradbury & Hewison, 1994; Munro, 1995), particularly since these involve face-to-face play, where parents of infants having early lip repair may find it easier to respond to infant social cues. Murray et al. (2008) added that any enhanced responsiveness in parents of early repair infants may have benefits for infant development.

In this regard, Murray et al. (2008) observed that mothers of infants having late repair were less positively involved with their infant, and the infant also spent less time looking at their mother at two months. Surprisingly, at six months and at twelve months, there were no differences on the mother-infant relationship between the groups (early repair, late repair and controls), and there was not an effect of antenatal diagnosis.

Additionally, in the same study, mothers of more disfigured infants were less positively involved, and infants with both cleft lip and palate looked less at their mother in early and late repair groups. Besides, Murray et al. (2008) found that infants having late lip repair surgery performed significantly more poorly cognitive development at 18 months old (assessed by Bayley Scales of Infant Development, 1993) than infants with no clefts, whereas infants having early repair were comparable to controls. These findings support the fact that having a disfigured infant may adversely affect maternal mental health, which in turn may disturb the mother-infant relationship, especially prior to surgery. In turn, reduced sensitivity to the infant in late repair group

mothers, at two months, mediated the adverse effect of late repair on infant cognitive outcome.

Similarly, Hentges et al. (2011) observed that school-aged children with CLP, particularly those with late lip repair, scored significantly lower than controls on cognitive development. Interestedly these group differences in verbal IQ were mediated by maternal sensitivity at 2 months of infants' age. This enhanced the importance of social interactions in the first months, not only for the socio-emotional but also for the cognitive infant development. The fact that less sensitive maternal interactions explained the group differences in infant cognitive outcome is consistent with the literature on the role of maternal behavior in influencing infant cognitive development in relation to infants with clefts (Speltz, Endriga, Hill, Maris, Jones, & Omnell, 2000; Wasserman & Allen, 1985).

These findings highlight the necessity to further explore the quality of parent-infant relationship, in the context of infant clefs, according to the timing of the repair surgery (early or late). In sum, parent–infant relationship is constantly and massively affected from birth. Based on these findings, and because almost all the literature that currently exists on this topic has focused on the mother-infant relationship, it results interesting to evaluate the quality of the parents–infant relationship in families with an infant with cleft lip and palate, taking into account the father's perspective.

In this regard, Zeanah et al. (1997) have suggested that, although observing infant–parent interactions is not equivalent to assessing the relationship, several domains of the infant–parent relationship can be observed in brief observations of dyadic interactions, including caregiver emotional availability, infant emotion regulation, caregiver support, security in the infant, and play (Lieberman & Zeanah, 1995;

Zeanah et al., 1997). In addition, the evaluation of psychological impact on both parents is an essential variable to take into account.

1.4. Psychological impact on parents of the CL/P malformation

Parenthood is a stressful period in itself, infants are in themselves a source of stress and parents can also be exposed to many other stressors. When there is a diagnosis of a congenital anomaly, parents must cope with the dual challenge of the transition to parenthood and of the prenatal or postnatal diagnosis, which is also associated with medical, financial, social and emotional demands (Fonseca et al., 2012).

Considering that the family provides the foundation of an infant's psychological and social development, the well-being of the parents represent a major protective factor regarding the infant's development. Parents naturally expect a perfect baby, without any problems. Thus, the detection of a cleft during pregnancy, or at birth, and the presence of a visible disfigurement may complicate the transition to parenthood and the encounter with the infant (Despars et al., 2011).

Parental reaction to the diagnosis varies greatly, but most parents express severe psychological shock. In addition, most of the general population has very little knowledge of the CLP malformation and what they do know is often biased by negative feelings (Rey-Bellet & Hohlfeld, 2004). Thus, parents of infant's with CLP are likely to experience emotional reactions such as confusion, denial, distress, and guilt (Bradbury & Hewison, 1994), as well as feelings of loss of control, helplessness, and even depression and anxiety (Rey-Bellet & Hohlfeld, 2004).

Besides, parents may feel damaged by their perceived incapacity to produce a healthy and typically formed baby who is free of physical defects (Dölger-Hafner, Bartsch, Trimbach, Zobel, & Witt, 1997;

Schlenker, Maury, Montoya, & Visier, 1998a; Schlenker, Montoya, Maury, & Visier, 1998b). As a result, parents have to initiates the grieving process necessary for the acceptance of their different child (Rey-Bellet & Hohlfeld, 2004).

Consequently, the announcement of a facial malformation affecting their baby is a major psychological burden for parents. The baby's presence becomes real and familiar as soon as it starts to move. While it is not yet born, he becomes a full member of the family, but the announcement of such a diagnosis during a prenatal ultrasound or at birth, the infant as he has been imagined disappears suddenly. His image is replaced with that of a stranger, and there is a sudden loss of the imagined baby, leading a major loss in their own ability to play their role in the inter-generational transmission (Grollemund et al., 2012b).

During the time of diagnosis and the perinatal period, parents usually feel strongly defenseless. Although, it is now widely accepted that the parents must be informed of any finding, and that prenatal diagnosis should be given to avoid the shock of discovery at birth and the feeling of being betrayed by the medical team (Rey-Bellet & Hohlfeld, 2004).

However, when, for different reasons, the diagnosis is only made at birth, it has been observed that the psychological impact on the parents is greater than when made prenatally (Bradbury & Hewison, 1994). Moreover, Davalbhakta and Hall (2000) found that parents who did not have the benefit of a prenatal diagnosis would have liked to be forewarned of the possibility of a malformation.

When the infant is born, parents of a baby with CL/P are brutally confronted with a damaged, split-open face, and the emotional burden of this event can limit their parental competences (Grollemund et al., 2012a). Sausse (1996) suggested that looking at the newborn's deformed face generates on parents contradictory emotions: distress,

horror, guilt, and desire to repair or to protect. In general, studies have shown that parents experience mental crisis in bringing up an infant with cleft lip and palate and in interacting with him or her (McWilliams, 1982; Turner, Thomas, Dowell, Rumsey & Sandy, 1997).

It is important to consider that the impact of the malformation will however be different depending on the type and extent of the CLP (Speltz, Armsden, & Clarren, 1990). Malformation, especially in the case of cleft lip that reach directly the face of the infant, may affect parents' attachment to the baby. Even after surgery, the remaining is the effect of an aggression, and this can be psychologically experienced as a stigma (Grollemund, Danion, Smaniotto, & Gruillot, 2011).

Apart from the emotional distress of having lost the perfect baby nurtured in their imagination, at birth and in the postnatal period, the parents have to cope with the anxiety and apprehension of nursing a cleft baby (Davalbhakta & Hall, 2000). As previously indicated, the treatment of cleft lip and palate is long, painful, highly complex, requiring extensive therapy to remediate speech, language, hearing, and feeding deficits.

In most cases, multiple surgeries are required to correct the associated anatomic deficits (Wellens & Vander-Poorten, 2006). Consequently, the level of care associated with orofacial clefts can result in a significant financial and psychological burden for families (McGrattan & Ellis, 2013). However, the specific impact of CL/P on the family has received little attention so far.

In accordance with the family stress and coping model (McCubbin, Thompson, & McCubbin, 1996), families of chronically ill children seek to restore balance and progress to a state of long-term adaptation. This adaptation is a process of reducing discrepancies between expectations and reality, and can be conceptualized as a continuum ranging from positive adaptation to negative adaptation. This model

takes into account several factors affecting the association between caring for a child with a medical condition and the parents' well-being. Thus, the present study aims to explore parental dimensions such as impact on the family, psychological distress, parental depression, and marital satisfaction reported by both mothers and fathers.

1.4.1. Impact on the family

Impact of the infant's medical condition on the family is defined as the parental perceptions of the effects of the infant's medical condition on the family (Stein & Reissman, 1980). In other words, any change in the normative behavior of the family which is directly attributable to the infant's medical condition, forcing adaptations in the family environment.

Most parents are emotionally shocked when confronted by the birth of a baby with a cleft lip and palate (CLP). Affected families may have to compensate for increased financial, social and personal impacts during the perinatal period and even childhood and adolescence. The diagnosis of a congenital malformation causes a crisis for the family. Parents inquiring about the etiology of the medical condition, and expectations about the future.

Additionally, the impact on the family can be in the form of increased burden and responsibility of caring for the infant at home for which families have varying physical and emotional resources. Besides, as most medical treatments are delivered during working hours, there is a potential for increased loss of income for the caregivers in the family leading to financial strain (Almesned, Al-Akhfash, & Al Mesned, 2013).

In this line, Stein and Reissman (1980) stated that infant's medical condition influences both structure and function of familial relationships in terms of financial burden, restrictions in social life, decreased

interaction with significant others, less time for other family members, and increased psychological distress.

According to Stein and Reissman (1980), financial burden is related to the economic consequences for the family; whereas social and familial impact refers to the disruption in normal social interaction within and outside the family system. Moreover, psychological distress is related to the personal disequilibrium experienced by the primary caretaker in relation to the infant's illness. Authors also found a dimension about the coping strategies employed by the family to master the stress of the medical condition, for example, talking and sharing, mutual support, and normalization of the infant's illness. Additionally, it is important to considerer that the disruption of each dimension (financial, social and familial impact, distress) in family functioning is not completely independent, since a family experiencing the impact in one area will be more likely to also experience the impact in other areas (Stein & Reissman, 1980).

Impact on the family was assessed in parents with an infant presenting different medical conditions, for example asthma (e.g., Hanson, Lapidus, Zuniga, & Murphy, 2000; Wood et al., 1993), epilepsy (e.g., Dehn et al., 2014), cancer (e.g., Alderfer & Kazak, 2006), and congenital anomalies (e.g., Albuquerque, Pereira, Fonseca, & Cavanavarro, 2012; Almesned, Al-Akhfash, & Al Mesned, 2013; Hunfeld et al., 1999). Besides, it has been found that the more severe and/or debilitating the illness, the greater the impact on the family (e.g., Kolk, Schipper, Hanewald, & Casari, 2000).

For instance, Wood et al. (1993) observed a moderate impact of infant's asthma on Hispanic parents. Likewise, Hanson et al. (2000) found a low impact on families with an infant presenting asthma, and this impact was not different between urban and rural parents nor by ethnicity.

Dehn et al. (2014) observed that younger age of the child presenting epilepsy was correlated to higher scores on parent's impact. Besides, a high seizure frequency had a higher impact on the family life. Also, more psychosocial problems of the child were associated with higher negative impact on the family. Finally, scores of impact on family were inversely correlated to the availability of support from friends.

Moreover, Alderfer and Kazak (2006) observed that parents of children with cancer often feel guilty in the diagnosis phase. Meeting with unfamiliar multidisciplinary professionals and receiving an immense load of information about the illness may overwhelm parents and increase their anxiety. These authors also found that the side effects of the cancer treatment caused anxiety and feelings of guilt in parents as they experienced their child suffering.

On the other hand, Almesned, Al-Akhfash, and Al Mesned (2013) conducted a study about social impact on families of children with complex congenital heart disease. They observed a high level of impact on the family because of the infant's heart disease. The highest impact perceived was in the familial/social burden, and the lowest impact was in the personal strain.

Similarly, in a study focused on the impact of congenital anomaly on the family, Albuquerque et al. (2012) observed that mothers reported higher scores on psychopathological symptoms and lower scores on quality of life than fathers. In addition, the perception of the severity of the congenital anomaly was significantly associated with paternal adjustment and with maternal burden. Interestingly, the timing of the diagnosis and the type of congenital anomaly were significantly associated with global and financial burden in both parents.

In a study about parental burden and grief in parents with a child with a congenital anomaly, Hunfeld et al. (1999) found that mothers felt significantly more personal strain than fathers. However, both parents

experienced the same low impact of financial burden and coping problems, and similar high limitations in social interactions due to the time they spend caring their infant. Interestingly, burden and grief were significantly associated with mothers, but no with fathers.

Furthermore, Hunfeld et al. (1999) observed that having a child with multiple -compared to isolated- congenital anomalies was associated with more burden and a low health status rating reported by mothers. Finally, these authors found that one year after the infant's birth, the mothers who had received a prenatal diagnosis reported significantly more total burden and social impact.

Concerning the impact of cleft lip and palate malformation on the family, Kramer, Beathge, Sinikovic and Schliephake (2007) conducted a study with families having an infant with a cleft aged between 6 and 24 months. They observed that, in general, the impact on coping strategies and on personal dimension was the highest, while financial impact and those affecting care of siblings were the lowest. Surprisingly, prenatal diagnosis of CL/P did not contribute to reduce the impact on family. On the contrary, social impact was increased in those families that were informed about the malformation before birth.

Moreover, Kramer et al. (2007) found significant differences related to the type of cleft. For example, families having an infant with cleft palate (CP) experienced higher general impact, financial impact, and social impact -in comparison to infants with cleft lip (CL) or cleft lip and palate (CLP)-, probably due to later surgery for reconstruction. On the other hand, families with a child with CL showed an increased impact on coping strategies. Besides, families with an infant having CLP experienced an increased impact on siblings. Interestingly, the laterality of the cleft (unilateral or bilateral cleft lip) had no relevant impact on the family.

Baker et al. (2009) found low levels of family impact on parents with a CLP child. They observed greater family impact in parents having a younger child or whose child had medical problems in addition to CLP. Moreover, parents who had avoidant-oriented coping strategies and perceived lower levels of support from friends and family reported higher family impact of CLP. Finally, parents who reported a greater family impact of CLP were those who had fewer confidants to talk to and were more likely to use emotional discharge and acceptance coping strategies.

According to the psychological impact of the waiting time prior to the first surgery, Slade, Emerson and Freedlander (1999) found no differences on psychological impact on parents depending on the timing of the infant's repair surgery. On the other hand, Galinier et al. (2008) suggested that early repair intervention is essential for the psychological status of the parents. Hence, findings are not conclusive and it is important to further study the psychological impact on parents depending the type and laterality of the cleft lip and palate malformation, and the timing of diagnosis and the repair surgery.

1.4.2. Psychological distress

Psychological distress is widely used as an indicator of the mental health. It is defined as a state of emotional suffering characterized by symptoms of depression (e.g., lost interest; hopelessness; sadness) and anxiety (e.g., restlessness; feeling tense) (Mirowsky & Ross 2002), that may impact on the social functioning and day-to-day living of individuals (Wheaton, 2007). Similarly, the concept of psychological distress refers to a general index of psychological alteration grouping together various affective and cognitive symptoms such as depression, anxiety or irritability (Ilfeld, 1976, 1978).

According to the stress-distress model, the defining features of psychological distress are the exposure to a stressful event that

threatens the physical or mental health, the inability to cope effectively with this stressor, and the emotional agitation that results from this ineffective coping (Horwitz, 2007; Ridner, 2004). This is to say, increased stress and poor adjustment may emerge when there is an imbalance between demands and coping resources, requiring substantial changes in family roles, goals, values, priorities and patterns of functioning (Baker et al., 2009).

Infant congenital anomalies and other medical conditions may be a source of psychological distress of parents and family. The first months after the birth of an infant with a congenital anomaly are particularly challenging and require individual and familial reorganizations to cope with the caregiving demands (Messias et al., 1995). Thus, psychological distress has been mainly used as the indicator of parental adjustment to their infant having a congenital anomaly (Fonseca, Nazaré, & Canavarro, 2012).

In this line, Viaux-Savelon et al. (2007) observed that, when there is a diagnosis of malformation, the infant-mother meeting at birth is tense, mothers are anxious, and this anxiety increases across the time. In the same way, Fonseca, Nazaré and Canavarro (2012) found that parents whose infants were diagnosed with a congenital anomaly showed higher levels of psychological distress than parents of healthy infants. Besides, they did not observe differences on psychological distress as a function of the timing of the diagnosis.

Similarly, it has been shown that after detection of a congenital anomaly, mothers experience significant acute psychological distress. Interestingly, gestational age predicted social dysfunction, health perception and acute psychological distress on mothers. The lowest level of psychological distress was in the group with the best prognosis and no ambiguity (Kaasen, et al., 2010).

Furthermore, Mazer et al. (2008) reported, in a research about the impact of a child with congenital anomaly on parents, that the level of fears and anxiety felt for the infant and its future appeared to decrease significantly over time for both parents. They suggested that these findings may be due to the fact that parents may have gained a better understanding of what to expect in the future, and because the acute severity of disease and the child's discomfort will usually have abated over time.

Likewise, Brosig et al., (2007) reported that parents having an infant with congenital heart disease experienced high levels of psychological distress at time of diagnosis and at birth, but they did not found differences either in mothers and fathers or in prenatal versus postnatal diagnosis. Six months after birth, the parental distress level was still higher in the prenatal diagnosis group, meanwhile the postnatal diagnosis group had distress level within the normal range.

In this regard, Fonseca, Nazaré and Canavarro (2012) observed that although higher levels of psychological distress were found in parents of infants with a congenital anomaly, one month after the disclosure of the diagnosis, studies suggest a gradual process of parental adjustment to the diagnosis (e. g., Lalor, Begley, & Galavan, 2009).

Particularly, in infant cleft lip and palate, mothers reported to experience less positive affect toward their baby having CLP, and more parental anxiety (Zeytinoglu & Davey, 2012). In addition, mothers described they feel worry about how to feed their infant, and they have feelings of loss and failure because they are unable to breastfeed. Moreover, parents may experience worry when they introduce their infant to others in their families, they are also worried about the ongoing care of the baby and the risk of cleft in future pregnancies (Collet & Speltz, 2007).

Psychological distress experimented by mothers may be also due to the appearance of the infant having a cleft lip. For instance, De Pascalis et al. (2017) found that mothers increased their visual attention towards their infant's mouth, from the first to the second month. However, the presence of a cleft lip was associated with decreased maternal gaze to the infant's mouth.

In this regard, a longitudinal study about the maternal life satisfaction and psychological distress after the birth of a child with a congenital anomaly conducted by Nes et al., (2014) showed that cleft lip and palate (CLP) had a temporary impact on maternal distress at 6 months postpartum. Moreover, higher life satisfaction and lower psychological distress was correlated with higher education and being a first-time mother. Besides, the timing of diagnosis appeared to have different impact on psychological distress after birth.

In addition, Stock and Rumsey (2015) reported that also fathers having an infant with CLP are at risk for psychological distress, especially during the first year. These findings were also confirmed by Zeytinoglu, Davey, Crerand and Fisher (2016a). However, in both studies the sample was formed by less than 20 fathers. Thus, it is important to further investigate psychological distress on mothers and fathers in order to have a more comprehensive perception of the psychological distress in parents having a cleft infant.

Additionally, it should be considered that perceptions about parenting experience may be different in mothers and fathers, because they tend to adopt different roles concerning the infant's care. For example, mothers tend to assume the role of main caregivers, whereas fathers tend to adopt the role of providers (Katz-Wise, Priess, & Hyde, 2010). Consequently, these different roles may lead to diverse perceptions about the demands.

Baker et al. (2009) found low levels of psychological distress in parents having a child with a cleft. Moreover, these authors observed that lower levels of perceived social support and a greater use of avoidance-oriented coping predicted more psychological distress. Similarly, parents who reported fewer confidants to talk showed higher levels of psychological distress. Moreover, Gray (2003) suggested that within the couple, the psychological distress for example, may impact their partner's adjustment and vice versa.

Literature previously described lead us to consider the importance of assessing psychological distress in mothers and fathers having an infant with cleft lip and palate, in terms of anxiety, irritability, cognitive problems, and depression during the first year after birth in order to provide a whole perspective of the psychological impact of the malformation on the family system.

1.4.3. Postnatal depression

The presence of depression in the postpartum period is a significant public health concern due to the morbidity it imposes upon women, their infants, and their families. Postnatal depression is a frequently observed condition and it varies greatly in different parts of the world. In Western cultures a prevalence of 10–15% during the first year postpartum has often been reported (e.g., Gavin et al., 2005). A systematic review reported that the point prevalence of major depressive disorder and minor depression ranged from 6.5–12.9% through the first 6 postpartum months, peaking at 2 and 6 months after delivery (Gavin et al., 2005).

Furthermore, the prevalence is even higher in women with a previous history of depression or with psychosocial risk factors such as low income or social isolation (Robertson, Celasun, & Stewart, 2003). Although the DSM-V (American Psychiatric Association, 2013) does not recognize postpartum depression as a separate diagnosis, patients

must meet the criteria for a major depressive episode and the criteria for the peripartum-onset specifier.

Commonly used classification systems (DSM-V, ICD-10) consider onset within four–six weeks postpartum, however, there are many studies supporting the occurrence of postnatal depression at six months or even a year postpartum (e.g., Gaynes et al., 2005; Howard, 2005; Josefsson, Berg, Nordin, & Sydsio, 2001; Musters, McDonald, & Jones, 2008). Furthermore, it has been seen that women diagnosed with postpartum depression are six times more likely to display recurrent depressive symptoms, as well as other physical and mental illnesses after 4 years compared with women without postpartum depressive symptoms (Josefsson & Sydsjo, 2007).

Numerous studies have contributed to a better understanding of maternal depression and its potentially adverse impact on infant development (e.g., Evans et al., 2012; Hall, 2012; Garner et al., 2012; Teti, Messinger, Gelfand, & Isabella, 1995; Weissman et al., 2006). For instance, it has been showed that maternal postnatal depression affects the quality of maternal care and predicts disturbances in children's later social, behavioral, cognitive, and physical development (e.g., Downey & Coyne, 1990; Goodman, Brogan, Lynch, & Fielding, 1993; Murray & Cooper, 2003; Rahman, Iqbal, Bunn, Lovel, & Harrington 2004). In this line, Bagner, Pettit, Lewinsohn and Seeley (2010) argued that the presence of maternal major depressive disorder during the first year postpartum represents a sensitive period and increases the risk of adverse infant's outcomes.

Research has shown that mothers with depression frequently have difficulties in scaffolding their infants' emotional needs. For example, Tronick and Reck (2009) highlight the impact of maternal depression on the infant affective state and on the capacity for repairing states of miscoordination. Infants of mothers with high levels of depressive

symptoms develop negative affective states that bias their interactions with others and exacerbate their affective problems. Further findings showed that male infants are more vulnerable than female infants to maternal depression (Tronick & Reck, 2009).

In addition, mothers with chronic depression have been observed to provide less social stimulation to their infants, including less touching, less talking and fewer games. These deficits in parenting skills have been correlated with a range of negative emotional developmental outcomes and represent important risk conditions for infants (Hernández-Reif, Field, Diego, Vera, & Pickens, 2005).

Although maternal depression has been widely studied, depressive symptoms in fathers has been less explored. A recent work has demonstrated that depression affects 5-10% of fathers, and a 10.4% in the prenatal and postnatal period (Paulson & Bazemore, 2010). Besides, men appear to show less depressive symptoms than women during pregnancy and immediate postpartum. It has been reported a peak between the 3th and 6th month after birth (25.6%), and a lowest rate in the first 3 months (7.7%) (Paulson & Bazemore, 2010).

It has been noted that men report some different symptoms when suffering with depression, with higher levels of irritability and expressions of anger (Madsen & Juhl, 2007). In this line, Wilson and Durbin (2010) observed that paternal depression may be manifested as a decrease in positive emotions, tenderness and sensitivity, and an increase in hostility and disengagement towards the infant. Ramchandani et al. (2005) found that children of depressed fathers during the postnatal period are at increased risk of behavioral problems at age 3.5 years, even when maternal depression and other risk factors had been controlled. This association was stronger in boys than in girls.

Similarly, in a different study, Ramchandani et al. (2011) observed that infants of depressed fathers showed higher distress during the first 3

months of life in comparison to infants of non-depressed fathers. Moreover, these authors found that paternal depression was associated with an increased risk of disharmony in partner relationships. Depressed men are more likely to express, and to elicit negative emotions in interactions with their partners, and also more likely to disengage from couple interactions at times of stress (Papp, Goeke-Morey, & Cummings, 2007).

Concerning the evaluation of parental depressive symptoms when the infant has a medical condition, Brehaut et al. (2009) observed more symptoms of depression in caregivers of children with medical problems, in comparison to caregivers of healthy children. In this regard, Solberg et al. (2011) observed that having an infant with a congenital heart disease had prolonged effects on mothers. They also found a significantly larger increase in depression and anxiety symptoms at 6 and at 18 months after birth.

In a study about the emotional reactions in parents having a child with congenital heart diseases, Bevilacqua et al. (2013) found that mothers were more depressed than fathers (45.7% vs. 20.0%). They did no observe significant differences between the prenatal and the postnatal diagnosis groups, but, in percentage, parents receiving prenatal diagnosis were more depressed, and those receiving postnatal diagnosis were more stressed.

Similarly, Fonseca, Nazaré and Canavarro (2013) found that mothers having an infant with a congenital anomaly presented higher levels of depressive symptoms than fathers. They suggest that these findings may be due to the greater number of changes that mothers should assume in the role of main caregiver of their infant.

Regarding parents having an infant with cleft lip and palate (CLP), Slutsky et al. (1969) found that a large percentage of parents experienced shock, hurt, disappointment, and helpless resentment, but

most mothers recover in short time. Besides, parents with a CLP infant had greater problems that those with a cleft palate (CP) infant.

Furthermore, Slade, Emerson and Freedlander (1999) found that depressive and anxiety symptoms were within the normal range, and that mother's emotional status improved significantly over the 6 months after birth. However, they did not find evidence to support the idea that an early surgical repair or at 3 months led to differential levels of anxiety or depressive symptoms. Nevertheless, mothers expressed a preference for an early surgery. Similarly, Weigl, Rudolph, Eysholdt and Rosanowski (2005) observed similar levels of depressive and anxiety symptoms between mothers with CL/P children and the control group (children with no clefts). However, in that study, only mothers of children older than 1 year took part.

Montirosso et al. (2012) observed that mothers of infants with cleft lip displayed more negative affectivity (i.e., depressive-like expressions) with a lower level of positive emotional expressiveness and enjoyment. Moreover, these mothers showed little effort to gain or maintain engagement with their infant, they smiled little and appeared tense during the interaction. Besides, the mother's facial expression and the tone of voice appeared sad and depressed. Additionally, Murray et al. (2008) found that mothers having an infant with a cleft lip reported higher depression levels than those in the control group.

Leemreis et al. (2014) said that parents of disfigured infants not only have to face hospital stays and medical treatment, but also, they have to cope with negative social feedback concerning the infant's appearance. This could increase the parental stress level and result in anxiety or depression. Thus, the disfigurement or an impairment of the infant may influence the emotional well-being of their parents, and in turn, parent's depression or anxiety may negatively affect the development of the infant.

This is, parental depression bears long-term negative consequences for the children's development and increases susceptibility to psychopathology (Apter-Levy, Feldman, Vakart, Ebstein, & Feldman, 2013; Matijasevich et al., 2015; Murray et al., 2011; van der Waerden et al., 2015), however, this negative effects can impact other subsystems in the family, including the parental relationship and the family as a whole (e.g., Letourneau et al., 2012).

1.4.4. Marital satisfaction

The transition to parenthood has been identified as a period of increased risk for deterioration in marital quality (Cowan, Cowan, Heming, & Miller, 1991; Schultz, Cowan, & Cowan, 2006). During the perinatal period, exchanges between parents become more instrumental, and emotional exchanges directed to one another decrease. Parents are centered on the infant, on the care he or she has to receive, and the organization of everyday life (Cowan & Cowan, 1992; McHale, 2007).

This predominantly instrumental communication often reduces marital satisfaction, which can last a year or two, but which also can develop into a sustained conflict –especially if parents already had a conflicted relationship before the infant's pregnancy (e.g., Fearnley, Shapiro, Gottman, & Carrere, 2000).

Lewis (1988a) defined marital competence as partner's ability to communicate emotions openly, to express warmness to each other, and to resolve differences in an open manner, with satisfactory results for both. Additionally, marital competence is related to the capacity to adjust to parenthood (Lewis, 1988b). Therefore, dyadic adjustment is also conceptualized as a process rather than an unchanging state (Spanier, 1976).

Similarly, marital satisfaction is defined as "a subjective experiencing of one's own personal happiness and contentment in the marital relationship" (Hendrick & Hendrick, 1997, p. 57). Marital adjustment - as it is also named- is conceived as a process based on a high degree of agreement between the couple, a low frequency of conflicts and negative interaction, a high frequency of common activities, and few affective or sexual problems (Spanier, 1976). Thus, marital adjustment is an important factor of family mental system (Bowman, 1990).

Furthermore, the expectancies each parent develops toward the other as a parent also play a fundamental role in marital satisfaction (Favez, Frascarolo, Keren, & Fivaz-Depeursinge, 2009). If these expectations are not met once the infant is born, it could lead to a deterioration of the marital relationship, for example when mothers expect more participation from the father in the infant's care (McHale, Lauretti, Talbot, & Pouquette, 2002; Van Egeren, 2003).

Moreover, marital discord may be a salient risk factor for psychopathology in offspring (Zahn-Waxler et al., 1988). The worsening of the marital relationship increases the probability of disrupting the infant's ability to socially adaptation. For instance, parents in conflict do not provide to their infant the emotional security necessary for emotional regulation, thus, infant may intervene directly in the conflict to tone it down, or respond by withdrawal (e.g., Davies, Cummings, & Winter, 2004).

A partner's well-being and satisfaction highly depends on the other's well-being and satisfaction as well as on integration in the social environment (Bodenmann, 1997). Literature suggested that mothers of children with congenital craniofacial conditions, including CLP, experience greater stress, feel less competent, and report more marital conflict than control groups do (Speltz, Morton, Goodell, & Clarren, 1993).

Stressors related to the hospital visits, surgeries, financial burden, feeding problems and uncertainties about complications might generate relationship instability (Dale et al., 2012). In this line, satisfaction may be seen as an outcome of adaptation and as an important predictor of parent's ability to cope and adapt to the responsibilities of caring for their infant with a medical condition (e.g., Benson & Gross, 1989). In addition, a satisfying relationship and a supportive partner can function as a buffer against distress and hopeless (Murphy, Christian, Caplin, & Young, 2007).

In this regard, Dale et al. (2013) observed that having an infant with congenital heart disease was not associated with reduced relationship satisfaction on mothers. However, in that study all women in the cohort experienced reduced and decreasing relationship satisfaction. Besides, mothers of infants with congenital heart diseases did not report higher percentage of divorce or separation at 36 months postpartum, in comparison to the cohort.

In a systematic review, van Schoors, Caes, Alderfer, Goubert and Verhofstadt (2016) found that a decrease in marital satisfaction was reported during the first year after diagnosis of infant cancer by fathers (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1998) and mothers (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1998; Wijnberg-Williams, Van de Wiel, Kamps, & Hoekstra-Weebers, 2015). The highest level of dissatisfaction was during the 2 months after diagnosis (Yeh, 2002), and significantly higher levels of satisfaction after treatment completion (Brown et al., 1992). Van Schoors, Caes, Alderfer, Goubert and Verhofstadt (2016) concludes that most couples adapt well to the crisis of pediatric cancer in terms of emotional closeness, support, and marital satisfaction. However, most experience difficulties in the domain of sexual intimacy, and reports on conflict.

Concerning families having an infant with cleft lip and palate (CL/P), Speltz et al. (1990) evaluated how having a child born with a craniofacial issue (CL/P, CP, and healthy children between 1 and 3 years old) affected marital satisfaction. Mothers caring for a child born with CL/P or CP tended to report lower marital satisfaction compared to the matched healthy control group.

Similarly, Pelchat et al. (2003) evaluated marital distress among mothers and fathers of toddlers born with CL/P, Down syndrome, and toddlers who had no disabilities. Marital stress was significantly associated with fathers' increased levels of insensitivity toward children born with CL/P. Fathers who reported less marital distress tended to be more sensitive toward their children born with CL/P. However, the timing of the diagnosis (prenatal vs. postnatal) was not reported in this study. Moreover, it has been observed that parents reported that while caring for a child born with CL/P, they often forgot about each other; some misunderstandings took place, causing them to withdraw from each other (Pelchat et al., 2004).

In this line, Zeytinoglu, Davey, Crerand, Fisher, and Akyil (2016b) conducted a study to explore the experiences of couples caring for an infant with CL/P. These authors found six dominant themes: relationship growth, challenges, roles and responsibilities, sources of support, talking about cleft, and lesson learned. Most couples reported that raising a child born with CL/P made their relationship stronger because this process increased faith in their partnership. Moreover, all couples reported experiencing relational challenges while caring for their infant, especially during the first few months after birth. The main difference between the diagnosis groups (prenatal vs. postnatal) seems to be feelings of resentment among postnatal mothers regarding their partners' lack of involvement in childcare.

Besides, couples described the importance of getting educated about CL/P, and having a plan to work as a team. Concerning the timing of diagnosis, couples with marital distress tended to be those receiving the diagnosis at postnatal but also with more preexisting stressors (i.e., fertility treatments, financial issues, struggles with role distribution) (Zeytinoglu et al., 2016b).

Although raising an infant with CL/P can be challenging for parents, few studies have examined how it impacts couples. Literature reported that raising an infant born with CL/P has an impact on parent's relationship, thus, couples should be routinely assessed for psychological issues. In addition, marital satisfaction may be seen as an adaptation of the couple to their infant having CL/P. As a result, it may be act as a protective factor for the family as a whole facing a stressor such a CLP diagnosis. These findings highlight the necessity of further studying the marital satisfaction -during the first year postpartum- on parents having an infant with CL/P.

2. METHOD

2.1. Objectives

2.1.1. General objective

To describe the evolution of infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship, at 4 and at 12 months postpartum, in infants with Cleft Lip and Palate malformation and their parents.

2.1.2. Specific objectives

a) To explore whether statistically significant differences exist in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the type of malformation groups (CL or CLP), at 4 and at 12 months postpartum.

b) To explore whether statistically significant differences exist in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the laterality of the malformation groups (unilateral or bilateral), at 4 and at 12 months postpartum.

c) To explore whether statistically significant differences exist in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the type of diagnosis groups (prenatal or postnatal), at 4 and at 12 months postpartum.

d) To explore whether statistically significant differences exist in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the early and late surgery groups, at 4 and at 12 months postpartum.

2.2. Hypothesis

2.2.1. General hypothesis

There will be statistically significant differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between 4 months and 12 months evaluation.

2.2.2. Secondary hypothesis

a) There will be statistically significant differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the Cleft Lip malformation group (CL) and the Cleft Lip and Palate malformation group (CLP), at 4 and at 12 months postpartum.

b) There will be statistically significant differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the unilateral malformation group and the bilateral malformation group, at 4 and at 12 months postpartum.

c) There will be statistically significant differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the prenatal diagnosis group and the postnatal diagnosis group, at 4 and at 12 months postpartum.

d) There will be statistically significant differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the early surgery group and the late surgery group, at 4 and at 12 months postpartum.

2.3. Participants

The study included a multi-center non-probabilistic sample of infants affected with cleft lip (with or without cleft palate) and their parents enrolled during the period from July 2011 to November 2013. The four recruitment centers were *the Centre Compétent du CHU de Strasbourg*, *the Centre Référent des malformations crânio-maxillo-faciales rares du CHU de Lille*, the *Centre Référent des malformations rares de la face et de la cavité buccale de l'APHP de Paris,* and the *Compétence Centre du CHU de Nancy,* all of them in France.

All infants born with a cleft lip (with or without cleft palate) and followed at the recruitment centers were eligible for the study and were invited to participate. The non-inclusion criteria were infants born before 34 weeks of amenorrhea, infants whose birth weight was under 2.00 kg, placed in foster homes, whose parents showed insufficient level of French and/or were illiterate.

At the first evaluation, 145 infants and their parents were included in the study. However, at the second evaluation, 123 infants and their parents continued to participate. Hence, 15.2% of the sample was missing in the second assessment. The missing part of the sample may be due to different reasons: refusal to continue to participate by one of the infant's parents in the course of the study follow-up, the occurrence of a complication in the course of treatment and/or a serious illness requiring major specific treatment, an unexpected complication related with the surgical intervention, serious illness or death of one of the parents, the parents moving house outside the regions involved in the research.

2.3.1. Infants

At the first evaluation, 145 infants and their parents participated in this research. Concerning the characteristics of the infants, 69.7% (*n* = 101) were male, and 83.3% (*n* = 80) were born by natural birth. Moreover, fifty-five (40.1%) of the infants were the first born, and the remaining 59.9% had siblings. At birth, infants had a mean of 39.08 (*SD* = 1.69) weeks of gestation. It should be considered that 46 values were missing in this characteristic, equivalent to 31.7% of the sample.

The main descriptive statistics of the infants are showed in table 2.1. As can be seen, at birth the mean weight was 3.24 (*SD* = 0.46) kg., in a range of 2.15 to 4.37 kilograms. Nonetheless, 38 values were missing (26.2% of the sample). Additionally, the mean height at birth was 49.54 (*SD* = 2.43) centimeters, in a range of 41.00 to 55.00 centimeters. However, 52 values were missing in height at birth, equivalent to 35.9% of the sample.

At the first evaluation, infants had a mean age of 4.04 (*SD* = 0.65) months, in a range of 3.07 to 6.30 months. It should be noted that 10 values were missing, equivalent to 6.9% of the sample. At the second assessment, 123 infants and their parents continued to participate in the research. The mean age of infants, in the second evaluation, was 12.29 (*SD* = 1.24) months, in a range of 9.00 to 16.14 months (see table 2.1). It is noteworthy that, in the case of babies born prematurely, the corrected age was taken into account for the time of the evaluation.

Table 2.1. *Main descriptive statistics of the characteristics of the infants participating (n = 145)*

		Weeks of gestation at birth	Birth weight (kg.)	Birth height (cm.)	Age at first evaluation (months)	Age at second evaluation (months)
N	Valid	99	107	93	108	93
	Missing	46	38	52	37	52
Mean		**39.08**	**3.24**	**49.54**	**4.04**	**12.29**
Median		39.00	3.25	50.00	4.07	12.11
Mode		38	2.64[a]	51.00	4.08	12.11
Standard deviation		1.69	0.46	2.43	0.65	1.24
Skewness		-0.591	.035	-0.794	1.026	0.832
Std. Error skewness		0.243	0.234	0.250	0.209	0.233
Kurtosis		0.571	0.277	1.287	2.099	2.767
Std. Error Kurtosis		0.481	0.463	0.495	0.414	0.461
Minimum		34	2.15	41.00	3.07	9.00
Maximum		42	4.37	55.00	6.30	16.14
Percentile	25	38.00	3.00	48.00	11.46	48.00
	50	39.00	3.25	50.00	12.11	50.00
	75	40.50	3.52	51.00	12.90	51.00

[a] There are more than one mode value. The smallest value is displayed.

According to the distribution of the participants to the medical center, as can be seen in table 2.2, half of the sample belonged to the Medical Center in Paris, and 22.6% belonged to the Medical Center in Lille, both, the Reference Centers in Craniofacial malformations.

Table 2.2. *Distribution of the infants participating according to the medical center (n = 145)*

Medical center	Frequency	Valid percentage
Strasbourg	25	17.8
Lille	33	22.6
Paris	73	50.0
Nancy	14	9.6
Total	145	100.0

2.3.2. Type of malformation (Cleft Lip or Cleft Lip and Palate)

Concerning the type of malformation, table 2.3 shows that 80.3% of the infants had a cleft lip with cleft palate (CLP), vs. 19.7% of infants with a cleft lip only (CL). However, 28 values were missing, equivalent to 19.3% of the sample.

Table 2.3. *Distribution of participants according to the type of malformation*

Type of malformation	Frequency	Valid percentage
Cleft Lip	23	19.7
Cleft Lip and Palate	94	80.3
Total	117	100.00
Missing values	28	
Total		100.0

2.3.3. Laterality of the malformation (*unilateral or bilateral*)

Regarding the laterality of the malformation, table 2.4 shows that 79.5% of the infants had a unilateral malformation, and only 20.5% had a bilateral malformation. It is important to consider that 28 values were missing, equivalent to 19.3% of the sample.

Table 2.4. *Distribution of participants according to the laterality of the malformation*

Type of malformation	Frequency	Valid percentage
Unilateral	93	79.5
Bilateral	24	20.5
Total	117	100.0
Missing values	28	
Total	145	100.0

2.3.4. Type of diagnosis (prenatal or postnatal)

As regards the type of diagnosis of the malformation (cleft lip with or without cleft palate), 84.2% of the diagnosis was performed during the prenatal period, and 15.8% was diagnosed at birth. However, 25 values were missing, equivalent to 17.2% of the sample (see table 2.5).

Table 2.5. *Distribution of participants according to the type of diagnosis*

Type of diagnosis	Frequency	Valid percent
Prenatal	101	84.2
Postnatal	19	15.8
Total	120	100.0
Missing values	25	
Total	145	

In relation to the subgroup of parents who knew the diagnosis in the prenatal period, the mean of the weeks of gestation at the moment of the diagnosis was 23.08 (*SD* = 4.52). Table 2.6 shows that most of the families was informed of the malformation diagnosis at the 22nd week of gestation. Nonetheless, 53 values were missing, corresponding to 36.6% of the sample.

Table 2.6. *Weeks of gestation at the moment of the diagnosis*

Weeks of gestation	Frequency	Valid percentage
15	1	1.1
16	2	2.2
17	1	1.1
18	9	9.8
20	8	8.7
21	4	4.3
22	**30**	**32.6**
23	11	12.0
24	8	8.7
25	4	4.3
26	1	1.1
28	2	2.1
29	1	1.1
30	1	1.1
31	1	1.1
32	2	2.2
33	1	1.1
35	4	4.3
39	1	1.1
Total	92	100.0
Missing values	53	
Total	145	

Furthermore, table 2.7 shows that the diagnosis was done by the gynecologist in the majority of cases, by the radiologist in the second place, and finally, by the midwife. It should be noted that 38 values were missing, correspondent to 26.2% of the sample.

Table 2.7. *Professional who did the diagnosis*

Professional	Frequency	Valid percent
Gynecologist	67	62.6
Radiologist	27	25.2
Midwife	13	12.1
Total	107	100.0
Missing values	38	
Total	145	

2.3.5. Waiting time prior to the first surgical intervention

According to the surgical protocols, on average, parents and infants waited 86.79 (*SD* = 51.48) days between the infant's birth and the lip reconstruction surgery, in a range of 8 to 253 days. However, 33 values were missing, corresponding to 22.8% of the sample. Table 2.8 shows the descriptive statistics of this variable.

Table 2.8. *Main descriptive statistics of the waiting time prior the first surgery (in days)*

		Waiting time prior the first surgery
N	Valid	112
	Missing	33
Mean		**86.79**
Median		98.00
Mode		21
Standard deviation		51.48
Skewness		0.297
Std. Error skewness		0.228
Kurtosis		-0.229
Std. Error Kurtosis		0.453
Minimum		8
Maximum		253
Percentile	25	31.25
	50	98.00
	75	116.75

According to the time of the surgery, infants were distributed between the early surgery group (0-90 days), and the late surgery group (91-253 days). Table 2.9 shows the frequencies in each group.

Table 2.9. *Distribution of participants according to the waiting time prior to the first surgery*

Surgery group	Frequency	Valid percent
Early surgery (0-90 days)	49	43.8
Late surgery (more than 90 days)	63	56.2
Total	112	100.0
Missing values	33	
Total	145	

2.3.6. Parents

Regarding the characteristics of the parents, at the first assessment, 145 mothers and 145 fathers participated in the present study. Of all parents, 94.9% reported living in couple (4.8% missing values). As can be seen in table 2.10, the mean age of the mothers was 30.82 (*SD* = 5.13) years old, in a range of 17.01 to 42.81 years. However, 7 values were missing, equivalent to 4.8% of the sample. Additionally, the mean age for fathers was 33.38 (*SD* = 5.88) in a range of 18.53 to 49.19 years. Nonetheless, 8 values were missing corresponding to 5.5% of the sample.

Table 2.10. *Main descriptive statistics of the age of the parents*

		Mother's age (years, months)	Father's age (years, months)
N	Valid	138	137
	Missing	7	8
Mean		**30.82**	**33.38**
Median		30.54	33.35
Mode		29[a]	28[a]
Standard deviation		5.13	5.88
Skewness		-0.136	-0.062
Std. Error skewness		0.206	0.207
Kurtosis		-0.015	-0.056
Std. Error Kurtosis		0.410	0.411
Minimum		17.01	18.53
Maximum		42.81	49.19
Percentile	25	27.85	29.49
	50	30.54	33.35
	75	34.44	37.81

[a] There are more than one mode value. The smallest value is displayed.

According to the professional activity, 71.7% of the mothers (5.5% of missing values) and 93.8% of the fathers (6.2% of missing values) reported to have a job. As can be noted in table 2.11, the majority of the mothers and the fathers reported to work as an employee. Only 13.1% of the mothers worked as an executive, in comparison with 22.2% of the fathers. Moreover, any father reported to be a housekeeper compared with 27.7% of mothers.

Table 2.11. *Professional activity of the parents*

Professional activity	Mother's frequency	Mother's valid percent	Father's frequency	Father's valid percent
Executive	18	13.1	28	22.2
Self-employed	1	0.7	7	5.6
Employee	77	56.2	72	57.1
Laborer	3	2.2	18	14.3
Housekeeper	38	27.7	0	0
Other	0	0	1	0.8
Total	137	100.0	126	100.0
Missing values	8		19	
Total	145		145	

Table 2.12 shows the monthly income of the families participating in this study. As can be seen, the 43.6% of the sample reported to have a family monthly income between 1501€ and 3000€, which is in line with the net income for employees in France reported by the National Institute of Statistic of France (INSEE) in 2012 (1596€) and in 2013 (1612€). Additionally, 44.4% of the families informed to have a family monthly income superior to 3000€. Accordingly, in terms of monthly income, families that participated in this study were within the average of the French population.

Table 2.12. *Monthly income of the families participating (n=145)*

Monthly income	Frequency	Valid percent
Equal or less than 1500€	16	12.0
Between 1501€ and 3000€	58	43.6
Equal or more than 3001€	59	44.4
Total	133	100.0
Missing values	12	
Total	145	

2.4. Instruments

This section details the description of the measuring instruments that were used in this research to evaluate infant social withdrawal behavior (ADBB: Alarm Distress Baby Scale, and the m-ADBB: the modified version of the Alarm Distress Baby Scale), psychological impact on parents (IOFS: Impact On Family Scale; IDP: Psychological Distress Index; EPDS: Edinburgh Postnatal Depression Scale; and DAS: Dyadic Adjustment Scale), and quality of the parent-infant relationship (PIPE: Pediatric Infant Parent Exam), at 4 and at 12 months postpartum.

It should be noted that, in the current study, the total score of all instruments has been used to assess the overall level of infant social withdrawal behavior, psychological impact on parents, and the quality of parent-infant relationship since the cut-off points are based on the total score, and because our study aimed to explore the overall level of these variables.

2.4.1. Alarm Distress Baby Scale (ADBB)

The ADBB scale was created by Guedeney and Fermanian (2001) as an instrument simple enough to be used effectively by professionals in clinical practice. The methodology is based on Winnicott's set *situation* (Winnicott, 1941), as well as on the NBAS (Brazelton, 1973) which both provide a given stimulation and observes the way the infant makes uses of it. The ADBB is a scale that therefore neither involves special apparatus nor a special sequence of prescribed interactions, and which does not require the parent's active interaction with the infant. One advantage of assessing an infant's social behavior with a relative stranger, rather than with his parent, is that it does not put the parent under any perceived pressure.

In this line, the ADBB scale was created for use in clinical practice settings in order to facilitate a more structured observation of social behavior, in infants from 2 to 24 months old, in the context of a routine physical exam. The assessment of social withdrawal is appreciated by the infant's reaction to stimulation, and pediatric consultation presents various stimuli in a short period of time and with a sufficiently identical sequence. Additionally, it is important that the clinician attempts to socially engage the infant –by talking, smiling, and touching-. The rating is done immediately after observation in a live situation or on videotape by a clinician trained in the scale.

The ADBB scale consists of eight items concerning the behavior and features of the infant: 1) facial expression, 2) eye contact, 3) general level of activity, 4) self-stimulating gestures, 5) vocalizations, 6) response to stimulation, 7) relation between the infant and the clinician, and 8) attraction, defined as the ability of the infant to attract and maintain the clinician attention. The score for each item represents the observer's general perception about the infant's performance in each dimension. The observer –trained in ADBB scale- rates on a 5-point scale ranging from zero to four. For each item, zero represents best functioning or normality of the infant and four, severe abnormality.

Total score is computed by summing the scores from the 8 items, thus, total score can range from 0-32. Studies have indicated that a total score of 5 or more is thought to be deviant and a sign of distress in the infant (De Rosa et al., 2010; Guedeney & Fermanian, 2001; Facuri-Lopes, Ricas, & Cotta, 2008; Matthey et al., 2005; Oliver, Yulitta, & Guedeney, 2016; Puura et al., 2007). Hence, higher total score reflects higher level of social withdrawal. Accordingly, table 2.13 shows a total score classification suggested by Guedeney (2015).

Table 2.13. *ADBB cut-off points according to the total score (Guedeney, 2015)*

ADBB total score	Classifications
0-4	Normal
5-10	Some concern
>10	Significant concern

It is important to consider that the ADBB scale has been designed to avoid using items that show dramatic changes with development, as development is so intense within the firsts two years of life. However, social withdrawal is not assessed without a context, and therefore depends on the stimulation and on the environment. This means that assessment is made within an age frame, so that the reaction of the infant is compared to what is expected at this particular age (Guedeney, 2015).

Regarding the construct validity of the French version of the ADBB scale, an exploratory factor analysis performed by Guedeney and Fermanian (2001) found two different factors which are described in table 2.14. According to Guedeney and Fermanian (2001), reliability was satisfactory with good internal consistency for both subscales (Cronbach's alpha coefficient was .80 for the first subscale, and .79 for the second one) and for the global scale (Cronbach's alpha coefficient was .83). In our research, the total score has been used to assess the overall level of infant social withdrawal, since the cut-off points are based on the total score.

Table 2.14. *Dimensions of the ADBB scale (*Guedeney & Fermanian, 2001*)*

Factor 1: interpersonal dimension	Factor 2: non interpersonal dimension
2. Eye contact	1. Facial expression
3. General level of activity	5. Vocalizations
4. Self-stimulating gestures	6. Response to stimulation
7. Relationship	
8. Attraction	

Concerning the internal consistency analysis of the ADBB scale in the current study, it was found a Cronbach's alpha coefficient of .82 (95% *IC* .77 to .86; *n* = 145) in the first assessment. The mean scores for each item are presented in table 2.15. It can be noted that item 5 "vocalizations" obtained the highest mean (*M* = 0.45, *SD* = 0.68) and item 3 "general level of activity" had the lowest mean (*M* = 0.16, *SD* = 0.39).

Additionally, the analysis of internal consistency indicated that the elimination of any of the items did not improve Cronbach's alpha value. The values of the correlation coefficients between the item and the total score reached from .38 to .69.

Table 2.15. *Internal consistency of the ADBB scale in the first evaluation (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Facial expression	0.39	0.63	.697	.774
2. Eye contact	0.21	0.51	.548	.798
3. **General level of activity**	**0.16**	0.39	.425	.814
4. Self-stimulating gestures	0.17	0.49	.388	.817
5. **Vocalizations**	**0.45**	0.68	.428	.819
6. Response to stimulation	0.28	0.51	.569	.795
7. Relationship	0.33	0.58	.590	.791
8. Attraction	0.39	0.65	.695	.774

In the second assessment, the internal consistency of the ADBB scale was .73 (95% *IC* .65 to .79; *n* = 123). Table 2.16 shows the mean scores for each item. As can be seen, item 5 "vocalizations" obtained the highest mean (*M* = 0.38, *SD* = 0.58) and item 3 "general level of activity" had the lowest mean (*M* = 0.07, *SD* = 0.25). The analysis of internal consistency indicated that the elimination of the "vocalizations" item would increase the Cronbach's alpha. However, this item was maintained due to its theoretical relevance, and because methodologically, their elimination does not considerably improve the overall level of internal consistency of the scale.

The values of the correlation coefficients between the item and the total score fluctuated from .23 to .68. Item 5 "vocalizations" and item 6 "response to stimulation" were below .34, though, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.16. *Internal consistency of the ADBB scale in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Facial expression	0.29	0.49	.688	.642
2. Eye contact	0.13	0.38	.465	.698
3. **General level of activity**	**0.07**	0.25	.514	.704
4. Self-stimulating gestures	0.08	0.40	.364	.716
5. **Vocalizations**	**0.38**	0.58	**.239**	.761
6. Response to stimulation	0.12	0.33	**.326**	.722
7. Relationship	0.21	0.43	.417	.706
8. Attraction	0.26	0.48	.551	.677

2.4.2. Modified version-Alarm Distress Baby Scale (m-ADBB)

The m-ADBB is the modified version of the ADBB scale created by Matthey, Crncec, Hales and Guedeney (2013). This scale includes only five items: facial expression, eye contact, vocalizations, general level of activity, and relationship. In addition, the scoring is changed to three global levels: "0=Satisfactory, 1=Possible problem or 2=Definite problem" for each item. Scoring modification was based on the rating used by professionals in the New South Wales (Australia), in the routinely screen for developmental milestones. As a result, it was decided to use this scoring format for the m-ADBB so that it could be easily incorporated into the routine clinical checkups.

The design of the m-ADBB was based on the analyses of the data collected on the ADBB in a study conducted by Matthey, Guedeney, Starakis and Barnett (2005). The results in this study showed that item 4 (self-stimulating gestures) was too difficult on which to obtain sufficient interrater agreement with raters experienced at working with infants. This suggests that this item could potentially be rated inaccurately too frequently to allow for robust psychometric properties of the scale (Matthey et al., 2013).

In addition, item 8 (attraction) was highly correlated (>*0*.61) with six of the other items (i.e., facial expression: +0.71; Relationship: +0.77), indicating that it was not contributing sufficient unique information to the scale. This could be explained because infants who smile (facial expression) and talk (vocalizations) will almost always cause the clinician to be interested in them. The opposite also may be true. Thus, this item was removed. Similarly, items 3 and 6 (response to stimulation and general level of activity, respectively) were quite highly correlated (+0.63); consequently, these two items were combined into one item titled "Activity". Regarding age suitability, Matthey et al.

(2013) recommend the use of the m-ADBB from 3 to 18 months of age.

In relation to the psychometric properties, Hartley et al. (2010) found a good reliability of the m-ADBB (Cronbach's alpha = .80) in infants of HIV-infected mothers, aged 10-12 months. Guedeney et al. (2013) also found a Cronbach's alpha coefficient = .81, in infants at 18 months age.

Moreover, Matthey et al. (2013), comparing performance of m-ADBB with that of the full ADBB, indicated that one "definite problem" or two "possible problems" on the m-ADBB approximated the validated clinical full ADBB cut-off score of 5 or more. In this line, Guedeney et al. (2013) also recommend a cut-off of 2 for the m-ADBB, this is, total scores higher than 2 indicate social withdrawal. These authors argued that because of its simplified coding and scoring scheme, as compared to the original ADBB, the m-ADBB may well prove to be a more practical solution for evaluating withdrawal behavior in vulnerable populations.

Regarding the internal consistency for the m-ADBB scale in the present study, it was found a Cronbach's alpha coefficient of .71 (95% *IC* .64 to .79; *n* = 136) in the first assessment. The mean scores for each item are presented in table 2.17. It can be noted that item 4 "vocalizations" obtained the highest mean (*M* = 0.46, *SD* = 0.69) and item 2 "eye contact" and item 3 "general level of activity" had the lowest mean (*M* = 0.18, *SD* = 0.49; *M* = 0.18, *SD* = 0.41, respectively). Additionally, the analysis of internal consistency indicated that the elimination of any of the items did not improve Cronbach's alpha value. The values of the correlation coefficients between the item and the total score reached from .38 to .63.

Table 2.17. *Internal consistency of the m-ADBB scale in the first evaluation (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Facial expression	0.44	0.64	.636	.597
2. Eye contact	0.18	0.49	.492	.668
3. General level of activity	0.18	0.41	.389	.705
4. Vocalizations	**0.46**	0.69	.393	.716
5. Relationship	0.40	0.61	.525	.650

In the second evaluation, the internal consistency of the m-ADBB scale was .65 (95% *IC* .54 to .74; *n* = 123). Table 2.18 shows the mean scores for each item. As can be seen, item 4 "vocalizations" obtained the highest mean (*M* = 0.36, *SD* = 0.56) and item 3 "general level of activity" had the lowest mean (*M* = 0.07, *SD* = 0.25). The analysis of internal consistency indicated that the elimination of the "vocalizations" item would increase the Cronbach's alpha. However, this item was maintained due to its theoretical relevance.

Moreover, the values of the correlation coefficients between the item and the total score fluctuated from .28 to .65. Only item 4 "vocalizations" was below .34, though, these items were preserved due to its contribution to Cronbach's alpha coefficient, and its theoretical significance.

Table 2.18. *Internal consistency of the m-ADBB scale in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Facial expression	0.28	0.49	.650	.451
2. Eye contact	0.11	0.34	.344	.624
3. General level of activity	0.07	0.25	.411	.616
4. Vocalizations	**0.36**	0.56	.282	.690
5. Relationship	0.20	0.40	.461	.571

2.4.3. Pediatric Infant Parent Exam (PIPE)

The Pediatric Infant Parent Exam (PIPE) was developed by Fiese, Poehlmann, Irwin, Gordon and Curry-Bleggi (2001). It is an observational measure focuses on the reciprocal nature of dyadic interactions between parents and their infant, during the first year of life. The PIPE involves systematically observing parent and infant playing an interactional game together. It was designed as a screening tool that is quick to administer, easy to use in a variety of settings, and with no require testing materials. The PIPE explicitly focused on the interaction rather than on the individual characteristics of the infant or parents.

The PIPE consists of asking parents to play an interactional game such as peek-a-boo with their infant during the pediatric consultation, and prior to invasive medical procedures. If mothers indicate that their infant do not play peek-a-boo, they are asked to identify another game that the infant enjoys, such as a tickling or bouncing game. Thus, the instruction to parents is to play a few minutes with their baby as they do at home. This sequence is filmed. Subsequently, the interaction between the parents and their infant is observed and scored -by a trained observer- for degree of interactional reciprocity and positive affect at starting the game, keeping the game going and stopping the game.

Each of these segments of the game is scored on a scale of 0 to 5, with lower scores reflecting more favorable interaction patterns. A total score is calculated by summing the scores from the three segments of the game (starting the game, keeping the game going and stopping the game). Thus, the theoretical amplitude of the total score is 0 to 15. According to Fiese, Poehlmann, Irwin, Gordon and Curry-Bleggi (2001), the more adaptive score is characterized by easy engagement between parent and infant. The game is sustained through smooth

back and forth movement, and there is a gradual cool down at the end. On the other hand, in the more maladaptive score, the parent is either disengaged or intrusive, and infants respond with negative affect. The game persists despite the infant protest, and there is an abrupt ending to the game. Table 2.19 shows the scoring description for "starting the game".

Table 2.19. *Scoring description for "starting the game" item of PIPE (Fiese, et al., 2001).*

SCORE	DESCRIPTION
0	*EASY ENGAGEMENT.* Parent easily gets infant's attention and infant shows positive affect. (Ex: Infant's face may brighten to sound of parent's voice; parent may caress child).
1	*INFANT DIFFICULT TO ENGAGE.* Parent has to work to get infant's attention but infant's affect is positive or neutral. (Ex: Parent jiggles infant repeatedly; infant eventually looks or smiles).
2	*PARENT DISENGAGED.* Half-hearted attempt by parent to get infant's attention but infant remains positive or neutral. (Ex: Parent looks back and forth at pediatrician rather than child; infant looks toward parent with anticipation).
3	*PARENT DISENGAGE/INFANT PROTEST.* Half-hearted attempt by parent followed by mild infant protest or blank stare. (Ex: Parent looks towards pediatrician, infant fusses or looks somber).
4	*INTRUSIVE ENGAGEMENT/INFANT AVOIDS OR FLAT AFFECT.* Parent repeatedly pokes or prods infant to get infant's attention and infant turns head away, protests, or has flat affect. (Ex: Parent pokes finger at infant and continues to prod when infant turns face away or fusses).
5	*INAPPROPRIATE AND BIZARRE ENGAGEMENT.* Parent starts game before infant looks or may make bizarre statements outside of context of game. Infant stares or seems to be detached from parent. (Ex: Parent comments on world affairs in a bizarre manner; child looks around).

Fiese et al. (2001) identified three groups based on the total score of the PIPE. The first group includes scores <5, and it is labeled "highly adaptive". These interactions are characterized by easy engagement, reciprocity and playfulness between mother and infant. The second group, labeled "marginally adaptive" includes PIPE scores of 5-9, and interactions are characterized by occasional signs of maternal disengagement or intrusiveness and infant negative affect, although these signs did not dominate the interaction. The third group labeled "problematic", comprises total scores of 10 or more. These interactions were characterized by sustained maternal disengagement or

intrusiveness combined with infant negative affect. The use of the PIPE in French population was confirmed by Rochette and Mellier (2007).

Concerning the internal consistency of the PIPE in the sample of the present study, it was obtained a Cronbach's alpha coefficient of .82 (95% *IC* .76 to .87; *n* = 123) in the first evaluation. Table 2.20 shows the mean score for each item of the PIPE. The analysis revealed that the elimination of any of the items did not improve Cronbach's alpha value. In addition, the values of the correlation coefficients between the item and the total score range from .63 to .74.

Table 2.20. *Internal consistency of the PIPE in the first evaluation (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Starting the game	0.64	1.07	.686	.766
2. Keeping the game going	**0.78**	1.11	.749	.700
3. Stopping the game	0.76	1.19	.632	.822

In the second assessment of the parent-infant interaction, the analysis of the internal consistency of the PIPE scale revealed a Cronbach's alpha coefficient of .77 (95% *IC* .69 to .83; *n* = 118). Table 2.21 shows the mean score for each item of the PIPE. Moreover, the analysis of internal consistency indicated that the elimination of the item "stopping the game" would increase the Cronbach's alpha however, this item is preserved due to its theoretical relevance in the evaluation of the parent-infant interaction. In addition, the values of the correlation coefficients between the item and the total score range from .45 to .73. Only "stopping the game" was below .50.

Table 2.21. *Internal consistency of the PIPE in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Starting the game	0.45	0.84	.652	.643
2. Keeping the game going	**0.55**	0.81	.737	.552
3. Stopping the game	0.51	0.90	.457	.862

2.4.4. Impact On Family Scale (IOFS)

The IOFS is an instrument that measures the impact of medical infantile conditions on general family quality of life, originally created by Stein and Riessman in 1980. In the current research we used the French version of the IOFS developed by Boudas Jégu, Grollemund, Quentel, Danion-Grilliat and Velten (2013). It should be noted that Stein and Reissman (1980; p. 466) defined impact as "the effects of a child's illness on the family system". They argued that changes occur in the family because of illness, forcing adaptations in the family environment. The IOFS focus on any change in the normative behavior of the family which is directly attributable to the infant's illness. This scale aims to assess negative influence of the illness in terms of restrictions in social life, decreased interaction with significant others, increased psychological distress by the primary caretaker, and less time for other family members.

Each item of the IOFS represents a certain statement about having a chronically ill or disabled child. The original version consisted of 33 items, which were assigned to five subscales and a total score: (1) Familial/Social Impact, e.g., "We see family and friends less because of the illness"; (2) Financial Burden, e.g., "The illness is causing financial problems for the family"; (3) Personal Strains, e.g., "Nobody understands the burden I carry"; (4) Mastery/Coping, e.g., "Because of what we have shared, we are a closer family"; (5) Impact on Siblings, e.g., "The school grades of my other children suffer because of the child's illness"; and Total score (without Impact on Siblings subscale).

The French version of the IOFS, adapted and validated by Boudas et al. (2013) in French-speaking parents with a child presenting a cleft lip (with or without cleft palate), comprises 15 items with one main dimension representing general negative impact on the social and

familial systems (see table 2.22). Parents are asked to rate in a 4 point Likert-type scale (from strongly disagree to strongly agree), according to the degree each of the items applies to the current situation of the family, with higher scores indicating higher psychosocial strains.

The total impact score is calculated by summing the results of all items, thus the total score ranging from 15 (minimum impact) to 60 (maximum impact). In order to obtain higher score values for greater impact, it is necessary to reverse the item values attributed (1 becoming 4, 2 becoming 3, etc.) in all items. As a result, the higher total score, the greater impact on the family. Boudas et al. (2013) reported a median score on the IOFS of 17 with an inter-quartile range of 15–24, and the analysis of the internal consistency yielded a Cronbach's alpha of .93.

Table 2.22. *Fifteen items conforming the French version of the Impact on Family Scale*

Items
1. Can't travel out of city
2. People treat us special
3. Little desire to go out
4. Hard to find reliable person to care for child
5. Need to change plans at last minute
6. See family and friends less
7. Wonder whether to treat child "specially"
8. Think about not having more children
9. No time for other family members
10. Family gives up things
11. Fatigue is a problem
12. Live from day to day
13. Nobody understands the burden
14. Travel to hospital is a strain
15. Live on roller coaster

Regarding the analysis of the internal consistency of the IOFS scale in the present study, a Cronbach's alpha coefficient of .86 (95% *IC* .82 to .89; *n* = 137) was found in the first evaluation for mothers. Table 2.23 shows the mean score for each item. As can be observed, item 14 "Travel to hospital is a strain" obtained the highest mean (*M* = 2.09,

SD = 0.91), and item 6 "See family and friends less" had the lowest mean (*M* = 1.26, *SD* = 0.55).

The analysis of internal consistency showed that the elimination of any of the items did not improve Cronbach's alpha value. Besides, the values of the correlation coefficients between the item and the total score range from .33 to .69, only item 7 was below .34, though, this item was preserved due to its contribution to Cronbach's alpha coefficient, and its theoretical significance.

Table 2.23. *Internal consistency of the IOFS for mothers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Can't travel out of city	1.31	0.58	.537	.856
2. People treat us special	1.47	0.69	.412	.861
3. Little desire to go out	1.36	0.69	.657	.850
4. Hard to find reliable person to care for child	1.88	0.94	.502	.857
5. Need to change plans at last minute	1.53	0.74	.561	.854
6. See family and friends less	**1.26**	0.55	.593	.854
7. Wonder whether to treat child "specially"	1.70	0.84	**.330**	.866
8. Think about not having more children	1.68	0.99	.386	.865
9. No time for other family members	1.55	0.72	.563	.854
10. Family gives up things	1.39	0.60	.656	.851
11. Fatigue is a problem	1.48	0.69	.692	.848
12. Live from day to day	1.51	0.80	.393	.862
13. Nobody understands the burden	1.74	0.78	.612	.851
14. Travel to hospital is a strain	**2.09**	0.91	.451	.860
15. Live on roller coaster	2.03	0.95	.532	.856

In the second assessment, the analysis of the internal consistency of the IOFS assessed on mothers yielded a Cronbach's alpha coefficient of .85 (95% *IC* .80 to .88; *n* = 119). Table 2.24 shows the mean score for each item. As can be noted, item 14 "Travel to hospital is a strain" obtained the highest mean (*M* = 1.88*, SD* = 0.90), and item 1 "Can't travel out of city", item 3 "Little desire to go out" and item 10 "Family gives up things" presented the lowest means (*M* = 1.16, *SD* = 0.43; *M* = 1.16, *SD* = 0.39 and *M* = 1.16, *SD*=0.39, respectively).

The analysis of internal consistency indicated that the elimination of any of the items did not improve Cronbach's alpha value. Moreover, the values of the correlation coefficients between the item and the total score range from .25 to .73. Item 7 and item 8 were below .34, though, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.24. *Internal consistency of the IOFS for mothers in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Can't travel out of city	**1.16**	0.43	.566	.839
2. People treat us special	1.24	0.49	.520	.840
3. Little desire to go out	**1.16**	0.39	.580	.840
4. Hard to find reliable person to care for child	1.70	0.84	.384	.848
5. Need to change plans at last minute	1.39	0.69	.547	.837
6. See family and friends less	1.18	0.40	.650	.837
7. Wonder whether to treat child "specially"	1.46	0.80	**.254**	.855
8. Think about not having more children	1.61	0.94	**.301**	.856
9. No time for other family members	1.36	0.61	.533	.838
10. Family gives up things	**1.16**	0.39	.707	.836
11. Fatigue is a problem	1.24	0.52	.735	.830
12. Live from day to day	1.41	0.67	.405	.845
13. Nobody understands the burden	1.57	0.78	.560	.836
14. Travel to hospital is a strain	**1.88**	0.90	.516	.839
15. Live on roller coaster	1.83	0.95	.655	.829

Concerning the fathers, the analysis of the internal consistency of the IOFS yielded a Cronbach's alpha coefficient of .86 (95% IC .82 to .89) in the first assessment. Table 2.25 shows the mean score for each item. As can be observed, item 14 "Think about not having more children"

obtained the highest mean (*M* = 2.16, *SD* = 0.91), and item "6 Live from day to day" presented the lowest mean (*M* = 1.26, *SD* = 0.54).

The analysis of internal consistency pointed out that the elimination of any of the items did not improve Cronbach's alpha value. In addition, the values of the correlation coefficients between the item and the total score range from .31 to .73, only item 7 was below .34, however, this item was preserved due to its theoretical relevance and its contribution to Cronbach's alpha coefficient.

Table 2.25. *Internal consistency of the IOFS for fathers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Can't travel out of city	1.35	0.63	.350	.858
2. People treat us special	1.55	0.68	.354	.858
3. Little desire to go out	1.40	0.70	.584	.847
4. Hard to find reliable person to care for child	1.86	0.93	.476	.854
5. Need to change plans at last minute	1.48	0.71	.614	.846
6. See family and friends less	**1.26**	0.54	.518	.852
7. Wonder whether to treat child "specially"	1.63	0.87	**.319**	.862
8. Think about not having more children	1.43	0.75	.467	.853
9. No time for other family members	1.59	0.68	.561	.848
10. Family gives up things	1.38	0.57	.582	.849
11. Fatigue is a problem	1.45	0.61	.734	.842
12. Live from day to day	1.51	0.80	.502	.851
13. Nobody understands the burden	1.60	0.73	.624	.845
14. Travel to hospital is a strain	**2.16**	0.91	.433	.856
15. Live on roller coaster	1.93	0.93	.558	.848

The analysis of the internal consistency of the IOFS, evaluated on fathers in the second assessment, obtained a Cronbach's alpha coefficient of .85 (95% *IC* .80 to .89). Table 2.26 shows that item 14 "Travel to hospital is a strain" obtained the highest mean (*M* = 1.82, *SD* = 0.88), and item 1 "Can't travel out of city" (*M* = 1.16, SD=0.40), item 3 "Little desire to go out" (*M* = 1.16, *SD* = 0.37), item 5 "Need to change plans at last minute" (*M* = 1.16, *SD* = 0.40), and item 10 "Family gives up things" (*M* = 1.16, *SD* = 0.37) presented the lowest means.

The analysis of internal consistency revealed that the elimination of any of the items did not improve Cronbach's alpha value. Additionally, the values of the correlation coefficients between the item and the total score range from .34 to .76.

Table 2.26. *Internal consistency of the IOFS for fathers in the second assessment (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Can't travel out of city	**1.16**	0.40	.343	.850
2. People treat us special	1.23	0.47	.485	.844
3. Little desire to go out	**1.16**	0.37	.516	.844
4. Hard to find reliable person to care for child	1.62	0.86	.500	.844
5. Need to change plans at last minute	**1.16**	0.40	.487	.845
6. See family and friends less	1.17	0.40	.465	.846
7. Wonder whether to treat child "specially"	1.51	0.75	.419	.848
8. Think about not having more children	1.44	0.81	.503	.843
9. No time for other family members	1.34	0.54	.766	.830
10. Family gives up things	**1.16**	0.37	.634	.841
11. Fatigue is a problem	1.24	0.43	.685	.837
12. Live from day to day	1.38	0.65	.421	.847
13. Nobody understands the burden	1.52	0.72	.559	.839
14. Travel to hospital is a strain	**1.82**	0.88	.493	.845
15. Live on roller coaster	1.68	0.82	.496	.844

2.4.5. Psychological Distress Index (IDP)

The IDP is a 14-item self-administered questionnaire which evaluates frequency of specific feelings and symptoms of psychological distress over the past week. That is to say, the occurrence of feelings of anxiety, depression, impaired cognition, and irritability. The IDP has been validated in both French and English for construct, criterion, and predictive validity (Préville, Boyer, Potvin, Perrault & Légaré, 1992). Studies have reported a Cronbach alpha coefficient of .89 (e.g., Bellerose et al., 1995).

In the present research, the psychological distress was evaluated with the IDP questionnaire with a dichotomous response scale indicating the agreement (2) or disagreement (1) with the sentence. This is to say, participants must rate each item referring if the feeling or symptom

has occurred in the week preceding the assessment, or not. The total score is calculated by summing the scores from the items. Thus, total score ranging from 14 to 28. High scores correlating with high psychological distress.

Moreover, items are grouped in four dimensions: anxiety, depression, irritability and cognitive difficulty, which theoretically are signs of psychological distress (Ilfeld, 1976, 1978). Table 2.27 shows the items conforming the depression dimension of the IDP. It is important to consider that in the current research, the total score has been used to assess the overall level of psychological distress, since the cut-off point is based on the total score.

Table 2.27. *Items conforming the depression dimension of the IDP*

Items
Did you feel hopeless about the future?
Did you feel lonely?
Did you feel downhearted or blue?
Did you feel bored or have little interest in things?
Did you cry easily or feel like crying?

The analysis of the internal consistency of the IDP scale in the sample of the present study, yielded a Cronbach's alpha coefficient of .79 (95% *IC* .73 to .83; *n* = 135) for mothers in the first assessment. Table 2.28 shows the mean score for each item. As can be observed, item two, referred to feel fearful or afraid, obtained the highest mean (*M* = 1.62, *SD* = 0.49), and item 7, related to have little interest in things, had the lowest mean (*M* = 1.09, *SD* = 0.29).

The analysis of internal consistency revealed that the elimination of any of the items did not improve Cronbach's alpha value. Additionally, values of the correlation coefficients between the item and the total score range from .22 to .59. Items 4, 7, 10, 13 and 14 obtained a correlation coefficient between the item and the total score inferior to

.34, however, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.28. *Internal consistency of the IDP for mothers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Did you feel tense or under pressure?	1.53	0.50	.563	.762
2. Did you feel fearful or afraid?	**1.62**	0.49	.431	.775
3. Did you feel nervous or shaky inside?	1.56	0.50	.537	.764
4. Did you feel hopeless about the future?	1.10	0.30	**.294**	.785
5. Did you feel lonely?	1.21	0.41	.430	.775
6. Did you feel downhearted or blue?	1.38	0.49	.435	.774
7. Did you feel bored or have little interest in things?	**1.09**	0.29	**.252**	.788
8. Did you cry easily or feel like crying?	1.61	0.49	.358	.782
9. Did you lose your temper?	1.47	0.50	.392	.779
10. Did you feel critical of others?	1.14	0.35	**.328**	.783
11. Did you feel easily annoyed or irritated?	1.51	0.50	.597	.758
12. Did you get angry over things that are not too important?	1.47	0.50	.488	.769
13. Did you have your mind go blank?	1.18	0.38	**.228**	.790
14. Did you have trouble remembering things?	1.22	0.42	**.281**	.787

Furthermore, the analysis of the internal consistency of the IDP scale, in the second evaluation of mothers, yielded a Cronbach's alpha coefficient of .80 (95% *IC* .75 to .85; *n* = 119). Table 2.29 shows the mean score for each item. As can be noted, item 11 referred to irritability, obtained the highest mean (*M* = 1.55, *SD* = 0.50) and item 4, related to feel hopeless about the future, had the lowest mean (*M* = 1.08, *SD* = 0.27).

The analysis of internal consistency indicated that the elimination of any of the items did not improve Cronbach's alpha value. In addition, the values of the correlation coefficients between the item and the total score range from .32 to .59. Only items 4 and 13 presented a correlation coefficient between the item and the total score below .34, though, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.29. *Internal consistency of the IDP for mothers in the second assessment (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Did you feel tense or under pressure?	1.50	0.50	.594	.779
2. Did you feel fearful or afraid?	1.50	0.50	.354	.800
3. Did you feel nervous or shaky inside?	1.50	0.50	.528	.785
4. Did you feel hopeless about the future?	**1.08**	0.27	**.325**	.801
5. Did you feel lonely?	1.23	0.42	.412	.795
6. Did you feel downhearted or blue?	1.39	0.49	.595	.779
7. Did you feel bored or have little interest in things?	1.16	0.37	.382	.797
8. Did you cry easily or feel like crying?	1.45	0.50	.356	.800
9. Did you lose your temper?	1.48	0.50	.348	.801
10. Did you feel critical of others?	1.21	0.41	.486	.789
11. Did you feel easily annoyed or irritated?	**1.55**	0.50	.564	.782
12. Did you get angry over things that are not too important?	1.52	0.50	.350	.800
13. Did you have your mind go blank?	1.17	0.38	**.334**	.800
14. Did you have trouble remembering things?	1.22	0.42	.354	.799

Concerning the evaluation of psychological distress on fathers, the analysis of the internal consistency in the first evaluation of fathers, revealed a Cronbach's alpha coefficient of .82 (95% *IC* .77 to .86; *n*=123). Table 2.30 shows that item 2, referred to feel fearful or afraid, obtained the highest mean (*M* = 1.50, *SD* = 0.50) and item 5, related to feel lonely, presented the lowest mean (*M* = 1.08, *SD* = 0.27).

The analysis of internal consistency showed that the elimination of any of the items did not improve Cronbach's alpha value. In addition, the values of the correlation coefficients between the item and the total score range from .32 to .67. Only items 9 and 13 had a correlation coefficient between the item and the total score lower than .34, though, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.30. *Internal consistency of the IDP for fathers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Did you feel tense or under pressure?	1.44	0.50	.628	.803
2. Did you feel fearful or afraid?	**1.50**	0.50	.373	.823
3. Did you feel nervous or shaky inside?	1.46	0.50	.675	.798
4. Did you feel hopeless about the future?	1.10	0.30	.523	.815
5. Did you feel lonely?	**1.08**	0.27	.375	.822
6. Did you feel downhearted or blue?	1.20	0.40	.545	.810
7. Did you feel bored or have little interest in things?	1.16	0.37	.446	.817
8. Did you cry easily or feel like crying?	1.25	0.44	.388	.821
9. Did you lose your temper?	1.35	0.48	**.326**	.826
10. Did you feel critical of others?	1.20	0.40	.475	.815
11. Did you feel easily annoyed or irritated?	1.41	0.49	.519	.812
12. Did you get angry over things that are not too important?	1.33	0.47	.489	.814
13. Did you have your mind go blank?	1.18	0.39	**.317**	.825
14. Did you have trouble remembering things?	1.19	0.39	.370	.822

In the second assessment of psychological distress of fathers, a Cronbach's alpha coefficient of .80 (95% IC .75 to .85; n=102) was obtained. Table 2.31 shows the mean score for each item. As can be observed, item 3, referred to feel nervous, obtained the highest mean (M = 1.38, SD = 0.49) and item 5, referred to feel lonely, presented the lowest mean (M = 1.04, SD = 0.20).

The analysis of internal consistency revealed that the elimination items 5, 8 and 13 would improve Cronbach's alpha value, however, these items are preserved due to its theoretical relevance. The values of the correlation coefficients between the item and the total score range from .23 to .62. As can be seen, items 5, 6, 8, 13 and 14 obtained a correlation coefficient between the item and the total score lower than .34, however, these items were preserved due to theirs contribution to Cronbach's alpha coefficient, and theirs theoretical significance.

Table 2.31. *Internal consistency of the IDP for fathers in the second assessment (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. Did you feel tense or under pressure?	1.35	0.48	.602	.782
2. Did you feel fearful or afraid?	1.31	0.47	.451	.796
3. Did you feel nervous or shaky inside?	**1.38**	0.49	.606	.781
4. Did you feel hopeless about the future?	1.07	0.25	.377	.802
5. Did you feel lonely?	**1.04**	0.20	**.126**	.813
6. Did you feel downhearted or blue?	1.16	0.37	**.334**	.804
7. Did you feel bored or have little interest in things?	1.13	0.34	.415	.799
8. Did you cry easily or feel like crying?	1.16	0.37	**.237**	.811
9. Did you lose your temper?	1.36	0.48	.536	.788
10. Did you feel critical of others?	1.15	0.36	.413	.799
11. Did you feel easily annoyed or irritated?	1.37	0.49	.621	.780
12. Did you get angry over things that are not too important?	1.39	0.49	.616	.780
13. Did you have your mind go blank?	1.12	0.32	**.232**	.810
14. Did you have trouble remembering things?	1.20	0.40	**.313**	.806

2.4.6. Edinburgh Postnatal Depression Scale (EPDS)

The Edinburgh Postnatal Depression Scale (EPDS) is a self-report designed for using in the course of routine contacts in order to enhance the detection of depressed mothers in the perinatal period. The original version was created by Cox, Holden and Sagovsky in 1987. In the current study, we used the French version developed by N. Guedeney and J. Fermanian in 1998. The EDPS has been used extensively in studies investigating mood disturbance in prenatal, postnatal and even beyond postnatal period (e.g., Cox, Chapman, Murray, & Jones, 1996; Murray & Carothers, 1990), in order to facilitate follow up assessment of the outcome of postnatal depression, which is important for the woman herself, but also to assess any adverse effects on the children (Murray, 1992). Thus, the EPDS is a screening tool for current depressive symptoms.

The EPDS consists of 10 self-report items, eight addressing depressive symptoms (e.g., self-blame, sadness), and two inquiring about anxiety symptoms (e.g., feeling scared or panicky). Each item is scored on a 4 point scale (from 0 to 3), according to the way mothers have felt during

the previous week. The total score of the EPDS is obtained by summing the scores from the items. In this procedure, it is required to recode the values in items: 3 and 5 to 10, this is, 0 becoming 3, 1 becoming 2, 2 becoming 1, and 3 becoming 0, with the aim of obtaining higher scores for more depressive symptoms. The minimum and maximum total score ranging to 0 to 30, respectively.

The validation of the French version of the EPDS was conducted by N. Guedeney and J. Fermanian (1998). These authors proposed a cut-off score of 10.5 as the best, with a good sensitivity (.80) and high specificity (.92). This is, total scores higher to 10.5 indicating depressive symptoms on mothers. Moreover, in the validation study of the French version of EPDS, the factor analysis produced two factors (see table 2.32). It is important to say that in our research, the total score has been used to assess the overall level of postnatal depression, since the cut-off point is based on the total score.

Table 2.32. Dimensions of the French version of EPDS (Guedeney & Fermanian, 1998)

Factor 1: Depressive symptoms	Factor 2: Depressive mood
Item 3 (guilty)	Item 1 (anhedonia)
Item 4 (anxiety)	Item 2 (anhedonia)
Item 5 (panic attacks)	Item 8 (sadness)
Item 6 (inability)	Item 10 (suicidal ideas)
Item 7 (sleep disorders)	
Item 9 (tearfulness)	

In the analysis of the internal consistency, Guedeney and Fermanian (1998) reported a Cronbach's alpha for the global scale of .76. In a different research in France, Rochette and Mellier (2007) suggested a cut-off of 10 for French sample. This is to say, a score equal or superior than 10 indicates the presence of current depressive symptoms.

Moreover, the EPDS is both reliable and valid to evaluate depressive and anxiety symptoms in fathers. Matthey, Barnett, Kavanagh and Howie (2001) found that when screening for a major or minor depression, a 9/10 is the optimum cut-off for men. Regarding the internal consistency, Matthey et al. (2001) found a Cronbach's alpha coefficient of .81 for men. In the present study, a cut-off of 10 is used for mothers and fathers.

The analysis of the internal consistency of the EPDS in the current research, a Cronbach's alpha coefficient of .81 (95% *IC* .76 to .86; *n* = 131) was obtained for mothers in the first evaluation. Table 2.33 shows the mean score for each item. As can be observed, item 3, referred to guilt, obtained the highest mean (*M* = 1.42, *SD* = 0.87) and item 10, related to suicidal ideas, presented the lowest mean (*M* = 0.15, *SD* = 0.50).

The analysis of internal consistency pointed out that the elimination of any of the items did not improve Cronbach's alpha value. Besides, the values of the correlation coefficients between the item and the total score range from .36 to .71.

Table 2.33. *Internal consistency of the EPDS for mothers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. I have been able to laugh and see the funny side of things	0.33	0.56	.484	.804
2. I have looked forward with enjoyment to things	0.30	0.58	.386	.812
3. I have blamed myself unnecessarily when things went wrong	**1.42**	0.87	.392	.814
4. I have been anxious or worried for no good reason	1.25	0.95	.493	.804
5. I have felt scared or panicky for no very good reason	0.81	0.88	.632	.785
6. Things have been getting on top of me	1.10	0.71	.364	.814
7. I have been so unhappy that I have had difficulty sleeping	0.55	0.83	.539	.796
8. I have felt sad or miserable	0.66	0.74	.653	.784
9. I have been so unhappy that I have been crying	0.55	0.80	.715	.776
10. The thought of harming myself has occurred to me	**0.15**	0.50	.368	.813

In the second evaluation of mothers, the analysis of the internal consistency of the EPDS yielded a Cronbach's alpha coefficient of .84 (95% *IC* .79 to .88; *n* = 118). Table 2.34 shows that item 3, referred to guilt, obtained the highest mean (*M* = 1.46, *SD* = 0.87) and item 10, related to suicidal ideas, presented the lowest mean (*M* = 0.13, *SD* = 0.44). The analysis of internal consistency revealed that the elimination of any of the items did not improve Cronbach's alpha value. In addition, the values of the correlation coefficients between the item and the total score range from .47 to .67.

Table 2.34. *Internal consistency of the EPDS for mothers in the second assessment (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. I have been able to laugh and see the funny side of things	0.39	0.69	.562	.825
2. I have looked forward with enjoyment to things	0.31	0.71	.492	.831
3. I have blamed myself unnecessarily when things went wrong	**1.46**	0.87	.503	.832
4. I have been anxious or worried for no good reason	1.12	0.91	.519	.830
5. I have felt scared or panicky for no very good reason	0.69	0.78	.483	.832
6. Things have been getting on top of me	0.98	0.75	.470	.833
7. I have been so unhappy that I have had difficulty sleeping	0.55	0.80	.646	.816
8. I have felt sad or miserable	0.73	0.81	.621	.819
9. I have been so unhappy that I have been crying	0.50	0.68	.671	.816
10. The thought of harming myself has occurred to me	**0.13**	0.44	.475	.835

Regarding the first evaluation of depression of fathers, the analysis of the internal consistency of the EPDS yielded a Cronbach's alpha coefficient of .80 (95% *IC* .75 to .85; *n* = 117). Table 2.35 shows the mean score for each item. As can be observed, item 3, related to guilt, obtained the highest mean (*M* = 1.09, *SD* = 0.97) and item 10, referred to suicidal ideas, showed the lowest mean (*M* = 0.14, *SD* = 0.49). The analysis of internal consistency indicated that the elimination of any of the items did not improve Cronbach's alpha value. Besides, values of the correlation coefficients between the item and the total score range from .34 to .66.

Table 2.35. *Internal consistency of the EPDS for fathers in the first assessment (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. I have been able to laugh and see the funny side of things	0.34	0.62	.344	.803
2. I have looked forward with enjoyment to things	0.28	0.61	.366	.801
3. I have blamed myself unnecessarily when things went wrong	**1.09**	0.97	.391	.807
4. I have been anxious or worried for no good reason	0.91	0.94	.539	.784
5. I have felt scared or panicky for no very good reason	0.58	0.81	.665	.766
6. Things have been getting on top of me	0.86	0.69	.582	.779
7. I have been so unhappy that I have had difficulty sleeping	0.52	0.79	.586	.777
8. I have felt sad or miserable	0.52	0.61	.624	.777
9. I have been so unhappy that I have been crying	0.15	0.50	.413	.798
10. The thought of harming myself has occurred to me	**0.14**	0.49	.416	.798

In the second assessment of the depression of fathers, the analysis of the internal consistency of the EPDS yielded a Cronbach's alpha coefficient of .83 (95% *IC* .78 to .88; *n* = 92). Table 2.36 shows that item 3, referred to guilt, obtained the highest mean (*M* = 0.98, *SD* = 0.92) and item 10, related to suicidal ideas, had the lowest mean (*M* = 0.10, *SD* = 0.42). The analysis of internal consistency showed that the elimination of any of the items did not improve Cronbach's alpha value. In addition, values of the correlation coefficients between the item and the total score range from .34 to .66.

Table 2.36. *Internal consistency of the EPDS for fathers in the second assessment (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1. I have been able to laugh and see the funny side of things	0.18	0.47	.514	.826
2. I have looked forward with enjoyment to things	0.20	0.56	.431	.831
3. I have blamed myself unnecessarily when things went wrong	**0.98**	0.92	.588	.819
4. I have been anxious or worried for no good reason	0.71	0.83	.658	.808
5. I have felt scared or panicky for no very good reason	0.46	0.78	.667	.807
6. Things have been getting on top of me	0.76	0.76	.649	.809
7. I have been so unhappy that I have had difficulty sleeping	0.32	0.61	.586	.817
8. I have felt sad or miserable	0.48	0.75	.509	.825
9. I have been so unhappy that I have been crying	0.11	0.31	.420	.835
10. The thought of harming myself has occurred to me	**0.10**	0.42	.347	.837

2.4.7. Dyadic Adjustment Scale (DAS)

The Dyadic Adjustment Scale was created by Spanier (1976) and it is currently the most widely utilized self-report measure of marital adjustment in the social and behavioral sciences. In the present study, we used the French version of the DAS developed by Antoine, Christophe and Nandrino (2008). The DAS often serves as a dependent measure of marital satisfaction (e.g., in marital therapy outcome studies, Christensen, Atkins, Berns, Wheeler, Baucom, & Simpson, 2004) or to classify "distressed" vs. "nondistressed" couples in marital interaction task research (e.g., Crane, Allgood, Larson, & Griffin, 1990). Moreover, the DAS is traditionally used to study the marital functioning, but nowadays, it is also used in different contexts in which the dyadic adjustment may be concerned, for example in psycho-oncology (Shields & Rousseau, 2004) and psychopathology studies (Whisman, Uebelacker, & Weinstock, 2004).

The DAS was created on the idea that marital adjustment is a process based on four components: a high degree of agreement between the couple (consensus), a low frequency of conflicts and negative interaction (satisfaction), a high frequency of activities in common (cohesion), and few affective or sexual problems (affective expression) (Spanier, 1976).

The French version of the Dyadic Adjustment Scale was developed by Antoine, Christophe and Nandrino (2008). Due to the divergence between the original internal structure and the factorial structure found in other studies, Antoine, Christophe and Nandrino (2008) decided to test the limits of the original DAS structure, and to identify a simple structure for the tool through exploratory factor analysis in French population.

As a result, sixteen items were preserved and organized according to two dimensions that explain 52% of the variance (see table 2.37). The

first factor, referred to the degree of agreement in the couple (DA), is formed by ten items, explaining 32% of the variance. The second factor, formed by six items, corresponds to the quality of the dyadic interaction (QI). This factor explains 20% of the variance. It should be noted that in the current study, the total score has been used to assess the overall level of marital satisfaction, since the cut-off point is based on the total score.

Table 2.37. *Fifteen items conforming the two dimensions of the French version of the Dyadic Adjustment Scale (Antoine, Christophe, & Nandrino, 2008)*

Degree of agreement (DA)	Quality of the interaction (QI)
1. Objectives	10. Exchange of ideas
2. Decisions	11. Discussions
3. Affection	12. Confidence
4. Friends	13. Common interest
5. Sexuality	14. Laugh together
6. Philosophy	15. Common project
7. Parents	
8. Divorce	
9. Nerves	
16. Happiness	

Participants complete the Dyadic Adjustment Scale appraising their marital satisfaction over the preceding 12 months. They should rate the extent of agreement or disagreement they have related to their partner in a six point Likert-type scale (from 1 strongly disagree to 6 strongly agree). The total score is obtained by calculating the sum of the items. In this process, it was necessary to reverse the values attributed in items 8 and 9 (1 becoming 6, 2 becoming 5, etc.), thus, high scores reflecting high adjustment in the couple. The theoretical range of the response scale is 16 to 96, with a cut-off of 54 suggested for French population (Antoine, Christophe, & Nandrino, 2008).

Moreover, Antoine, Christophe and Nandrino (2008) found that the factors were shown to be stable and similar for sex. Furthermore, the internal consistency was satisfactory, with a Cronbach's alpha coefficient of .89 for the total scale, .89 for the sub-scale of the degree

of agreement in the couple (DA), and .75 for the sub-scale of quality of the interactions (QI). Additionally, the DAS-16 was strongly correlated with the full DAS version (r = .97; p < 0.01), which confirms the possibility of score equivalence.

Regarding the analysis of the internal consistency of the DAS in the present study, a Cronbach's alpha coefficient of .87 (95% *IC* .84 to .90; n = 129) was obtained for the mothers in the first evaluation. Table 2.38 shows the mean score for each item. As can be observed, item 8, related to divorce, obtained the highest mean (M = 5.40, SD = 0.88) and item 15, referred to work in a common project, presented the lowest mean (M = 3.90, SD = 1.48).

The analysis of internal consistency revealed that the elimination of any of the items did not improve Cronbach's alpha value. As well, values of the correlation coefficients between the item and the total score range from .37 to .66.

Table 2.38. *Internal consistency of the DAS for mothers in the first evaluation (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1 Objectives	4.78	0.95	.526	.866
2 Decisions	5.03	0.95	.660	.861
3 Affection	4.89	1.08	.455	.869
4 Friends	4.72	1.10	.515	.866
5 Sexuality	4.48	1.15	.464	.869
6 Life Philosophy	4.47	1.16	.571	.864
7 Parents –in law	4.43	1.19	.443	.870
8 Divorce	**5.40**	0.88	.660	.862
9 Nerves	4.51	0.91	.504	.867
10 Exchange of ideas	4.03	1.02	.398	.871
11 Discussions	4.50	1.03	.638	.861
12 Confidence	4.87	1.17	.639	.860
13 Common interest	4.20	1.29	.470	.869
14 Laugh together	4.98	0.88	.570	.865
15 Common project	**3.90**	1.48	.376	.876
16 Happiness	4.75	0.93	.502	.867

The analysis of the internal consistency of the DAS in the second evaluation of mothers, yielded a Cronbach's alpha coefficient of .91

(95% *IC* .88 to .93; *n* = 105). Table 2.39 shows that item 8, related to divorce, obtained the highest mean (*M* = 5.31, *SD* = 1.00) and item 15, related to work in a common project, had the lowest mean (*M* = 3.87, *SD* = 1.54). The analysis of internal consistency showed that the elimination of any of the items did not improve Cronbach's alpha value. In addition, values of the correlation coefficients between the item and the total score range from .34 to .75.

Table 2.39. *Internal consistency of the DAS for mothers in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1 Objectives	4.70	0.88	.555	.907
2 Decisions	4.98	0.84	.553	.907
3 Affection	4.71	1.13	.643	.904
4 Friends	4.70	0.99	.721	.902
5 Sexuality	4.26	1.19	.532	.908
6 Life Philosophy	4.55	0.94	.639	.905
7 Parents –in law	4.50	1.26	.612	.905
8 Divorce	**5.31**	1.00	.752	.901
9 Nerves	4.49	0.98	.569	.907
10 Exchange of ideas	4.05	1.03	.349	.913
11 Discussions	4.57	1.06	.700	.903
12 Confidence	4.79	1.27	.626	.905
13 Common interest	4.05	1.30	.643	.904
14 Laugh together	4.74	0.90	.643	.905
15 Common project	**3.87**	1.55	.451	.914
16 Happiness	4.63	1.01	.751	.901

Concerning the evaluation of marital satisfaction of fathers, the analysis of the internal consistency of the DAS revealed a Cronbach's alpha coefficient of .87 (95% *IC* .83 to .90; *n* = 122) in the first assessment. Table 2.40 displays the mean score for each item. As can be noted, similarly to mothers, item 8, related to divorce, had the highest mean (*M* = 5.56, *SD* = 0.83) and item 10, referred to exchange of ideas, obtained the lowest mean (*M* = 3.84, *SD* = 1.05).

The analysis of internal consistency pointed out that the elimination of any of the items did not improve Cronbach's alpha value. Additionally, values of the correlation coefficients between the item and the total score range from .18 to .62. Only the item 10 obtained a correlation

coefficient between the item and the total score inferior than .34, however, this item was maintained due to its contribution to Cronbach's alpha coefficient, and its theoretical relevance.

Table 2.40. *Internal consistency of the DAS for fathers in the first evaluation (T0)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1 Objectives	4.78	0.93	.626	.859
2 Decisions	5.04	0.84	.560	.862
3 Affection	4.94	1.01	.509	.863
4 Friends	4.67	1.08	.536	.862
5 Sexuality	4.58	1.07	.551	.861
6 Life Philosophy	4.68	1.05	.647	.857
7 Parents –in law	4.53	1.16	.459	.865
8 Divorce	**5.56**	0.83	.532	.863
9 Nerves	4.68	0.95	.543	.862
10 Exchange of ideas	**3.84**	1.05	**.184**	.877
11 Discussions	4.59	1.01	.685	.855
12 Confidence	4.55	1.40	.564	.861
13 Common interest	4.14	1.18	.443	.866
14 Laugh together	4.93	0.97	.544	.862
15 Common project	4.07	1.56	.394	.873
16 Happiness	4.84	0.78	.582	.862

In the second evaluation of marital satisfaction of fathers, the analysis of the internal consistency of the DAS, a Cronbach's alpha coefficient of .90 (95% *IC* .87 to .92; *n* = 98) was obtained. In table 2.41 we can observe that item 8, related to divorce, had the highest mean (*M* = 5.54, *SD* = 0.81) and item 10, referred to exchange of ideas, presented the lowest mean (*M* = 3.89, *SD* = 1.05). The analysis of internal consistency showed that the elimination of any of the items did not improve Cronbach's alpha value. Moreover, the values of the correlation coefficients between the item and the total score range from .35 to .73.

Table 2.41. *Internal consistency of the DAS for fathers in the second evaluation (T1)*

Items	Mean	SD	Corrected Item-Total correlation	Cronbach's alpha if item deleted
1 Objectives	4.89	0.82	.569	.898
2 Decisions	5.05	0.90	.587	.898
3 Affection	4.86	1.08	.633	.896
4 Friends	4.74	0.99	.482	.901
5 Sexuality	4.39	1.18	.664	.894
6 Life Philosophy	4.57	1.12	.722	.892
7 Parents –in law	4.54	1.12	.498	.901
8 Divorce	**5.54**	0.81	.639	.897
9 Nerves	4.63	1.05	.570	.898
10 Exchange of ideas	**3.89**	1.05	.356	.905
11 Discussions	4.54	1.10	.716	.893
12 Confidence	4.46	1.29	.569	.899
13 Common interest	3.96	1.13	.659	.895
14 Laugh together	4.67	1.00	.656	.895
15 Common project	3.99	1.39	.366	.908
16 Happiness	4.78	0.83	.736	.894

In summary, internal consistency analyses of all instruments revealed that, according to George and Mallery (2003), Cronbach's alpha values were from acceptable to good for the sample participating in the current study. Only the m-ADBB in the second assessment showed an internal consistency labeled as questionable (.65). However, Nunnally (1967) states that, in exploratory studies, a Cronbach's alpha value from 0.6 to 0.5 may be considered acceptable.

This is to say, all instruments used in the current research have shown to be reliable in assessing infant social withdrawal behavior (ADBB and m-ADBB), psychological impact on parents (IOFS, IDP, EPDS and DAS), and the quality of parent-infant relationship in families having an infant with CL/P.

2.5. Procedure

The sample collection occurred from July 2011 to November 2013. In the course of the postnatal consultation, the surgical team proposed parents whose infants were born with a cleft lip (with or without cleft palate) to participate in the study. The presentation of the research protocol was done by the surgeons during the consultation following the birth of the infant, so that, all parents were informed in similar conditions. In this session, the interest, the objectives and the procedure of the research were explained to the parents. At the end of the consultation, parents who agreed to participate in the study signed a consent form for themselves and their infant (appendix 1).

The study took place during the first year after the infant's birth. A first evaluation was conducted at 4 months (T0), and a second evaluation at 12 months (T1) after birth. The choice of positioning T0 at 4 months was based on the following arguments: a) because of the variability of the protocols used by the surgical teams in each of the centers, it was not possible to define T0 and T1 that correspond to a specific examination for all the centers; b) at this age, the infants for whom the surgery occurs early have already had their operation. They can therefore be compared to infants whose operation occurred subsequently.

On the other hand, the choice of positioning the second evaluation (T1) at 12 months is justified because when the infant is one year old, this is sufficiently distant from both birth and the first surgical intervention to repeat the evaluations and questionnaires used in the first stage of the study. This would allow the comparison of psychological perceptions of the parents and the social development of the infant between T0 and T1, as well as to explore the question of the timing of surgery.

For each evaluation, parents were contacted and cited by the surgical team in their corresponding medical center. Before each appointment, a specifically developed questionnaire collecting socioeconomic variables and medical information was sent to the parents by mail. They completed this questionnaire at home and they hand it over at the appointment.

At T0 and T1, the infant was examined by a specially trained professional in order to evaluate the social withdrawal behavior, using the ADBB and the m-ADBB scales. At four months (T0), parents and their infant arranged an appointment in the Department of Pediatric Surgery in which the infant had been taken care of since birth. In the consultation, the infant was sitting in a baby chair in order to be available to interaction. The professional was placed at the same altitude as the infant, so that he tried to interact with the infant through the gaze, the smile, the talk and the touch. Different toys were shown one by one to the infant. Then, the baby was taken out of the chair and lay down on the mat, where the infant was placed on his stomach, on his back, and urged to turn by him or herself. Finally, the infant was put back in the chair. This procedure lasted between ten and fifteen minutes, and it was videotaped in order to subsequently assess social withdrawal behavior through ADBB and m-ADBB scales. It should be noted that all centers had the same baby chair, and the same toys.

Afterward, in the same consultation, parents were asked to play an interactional game such as peek-a-boo with their infant. If parents indicated that their infant do not play peek-a-boo, they were asked to identify another game that the infant enjoys, such as a tickling or bouncing game. Thus, the instruction to parents was to play a few minutes with the baby as they do at home. This sequence was filmed in order to successively evaluate the quality of the parent-infant relationship, through the PIPE scale.

Additionally, self-administered questionnaires were given to the parents to complete independently during the consultation, in order to evaluate the impact of the malformation on the family (IOFS), the parental psychological distress (IDP), the parental postnatal depression (EPDS) and the dyadic adjustment of the parental couple (DAS).

At 12 months (T1), parents and their infant agreed a second appointment at the Department of Pediatric Surgery. The evaluations of infant social withdrawal behavior -using the ADBB and the m-ADBB scales-, and the parent-infant relationship –through the PIPE scale-, were carried out. The procedures were identical to the previous one (T0), but the toys and games were adapted to the age of the infant. These sessions were also filmed. Likewise, parents were asked to complete the same questionnaires independently, during the consultation.

Subsequently, to evaluate social withdrawal behavior, videotapes of infant-clinical interaction were coded according to the ADBB manual (Guedeney, 2015), by two independent trained coders. One of the coders was the student author of this thesis (CPM), who was trained in clinical and research use of the ADBB and the m-ADBB scales by Dr. Antoine Guedeney. The second coder was an infant psychologist trained in ADBB scale (PG). The inter-rater agreement was good (*kappa* = .837; *kappa* = .879, at T0 and T1, respectively). Simultaneously, the student presenting this thesis (CPM) also scored social withdrawal through the m-ADBB scale (Matthey et al., 2013). In the current study, the ADBB and the m-ADBB scores obtained by the student author of this thesis (CPM) were used to data analysis.

Moreover, to evaluate the quality of the relationship, videotapes of parent-infant interaction was rated by a trained clinician through the PIPE scale (Fiese et al., 2001). Likewise, total scores for each

questionnaire (IOFS, IDP, EPDS and DAS) were calculated. The medical information about the precise type and laterality of CL/P, the time when the diagnosis occurred, and the timing of the first repair surgery was consulted in the medical records. All data were entered into the Statistical Package for the Social Sciences (SPSS) for the subsequent analysis.

Finally, it is important to point out that this study was approved by the *Comité de Protection des Personnes Est IV* of the Strasbourg Hospital on 18/11/2009. This approval is valid for all four of the study sites in France. The protocol conforms to the Helsinki Declaration and Good Clinical Practice guidelines of the International Conference on Harmonization. This trial is registered at ClinicalTrials.gov, Identifier: NCT00993993.

2.6. Design and data analysis

2.6.1 Variables

2.6.1.1. Independent variables. Grouping variables.

A) Type of malformation: Cleft Lip (CL, group 1) involves disruption of tissue planes above the lip, whereas Cleft Lip and Palate (CLP, group 2) includes the disruption of tissue planes above the lip extending into the palate (hard and/or soft) (Dixon et al., 2011). According to Corbo-Rodríguez and Torres (2001), more than 50% of cases includes Cleft Lip and Palate (CLP). Information about the type of malformation for each infant was obtained through the medical records.

B) Laterality of the malformation: The Cleft Lip and Palate malformation could be unilateral (group 1) or bilateral (group 2). This is to say, the disruption of tissue planes above the lip may affect only one side (unilateral) or both sides (bilateral) of the upper lip (Zeytinoglu et al., 2012). Information about the laterality of the

malformation for each infant was obtained through the medical records.

C) Type of diagnosis: The diagnosis of the Cleft Lip and Palate malformation could be performed either in utero (prenatal, group 1) or at birth (postnatal, group 2). Imaging techniques are now sufficiently fine to reveal a CL/P as early as 12 weeks into pregnancy, however, it may be the case that the malformation be discovered only at childbirth (Grollemund et al., 2012a; Habersaat et al., 2013). This information was obtained for each infant through the medical records.

D) Waiting time prior to the first surgery: Nowadays, the modalities of reconstructive surgery vary according to the severity of the malformation and the protocol followed by each surgical team. Certain teams prefer early intervention, this is immediately after birth; whereas other medical teams prefer to wait three of even six months. In the present research, infants were grouped in an early surgery intervention group (0-90 waiting days prior to the surgery, group 1) and a late surgery intervention group (more than 90 waiting days prior to the surgery, group 2). It is important to consider that the first surgery intervention is on the cleft lip, and it is the only surgery carried out during the first year of infant's life.

E) Time of the evaluations: The infant social withdrawal behavior, the psychological impact on parents and the quality of parent-infant relationship were assessed at four (T0, time 0) and at twelve months after infant's birth (T1, time 1). The choice of positioning T0 at 4 months was because of the variability of the protocols used by the surgical teams in each of the centers, it was not possible to define T0 and T1 that correspond to a specific examination for all the centers. Additionally, at this age, the infants for whom the surgery occurs early have already had their operation. On the other hand, the choice of positioning T1 at 12 months is justified because when the infant is one

year old, this is sufficiently distant from both birth and the first surgical intervention to repeat the evaluations used in the first stage of the study.

Moreover, it should be pointed out that at the time of knowing the diagnosis -whether in the prenatal stage or at birth- a psychologist provides parents with information about the CL/P malformation and solves the doubts they may have about it. Subsequently, the psychologist has an appointment with the parents during the first hospitalization (at the moment of the first surgery), in which he/she provides information and support for parents. Moreover, the medical team gives guidelines to parents for feeding the baby, as far as the post-surgical care. Afterwards, parents may have an appointment with the psychologist at their own request or at the demand of the medical team.

The information and psychological support that the parents receive are aimed to inform about the CL/P malformation and its treatment, as well as to resolve doubts in this regard. Therefore, it is not considered that there is a standardized psychological intervention from any specific theoretical model. This protocol is the same for all parents having an infant with cleft lip (with or without cleft palate) (CL/P).

2.6.1.2. Dependent variables

a) *Infant social withdrawal behavior:* It consists of diminished or lacking of either positive (e.g., smiling, eye contact, cooing) or negative behaviors (e.g., dampening of protest, and diminished crying) (Guedeney, 1997). Moreover, social withdrawal is considered a way of handling repetitive or durable violations of his or her expectations within parent-infant interactions (Murray & Trevarthen, 1985; Puura et al., 2010). This variable was operationalized through the ADBB scale (Alarm Distress Baby Scale) (Guedeney & Fermanian, 2001) and the short version m-ADBB (Matthey et al., 2013).

b) Quality of parent-infant relationship: Healthy interactions are characterized by parent and infant establishing joint attention, modulating, or matching voice and facial affect (Nicely, Tamis-LeMonda, & Grolnick, 1999), and gradually decreasing activity to end the interaction. Whereas maladaptive interactions are characterized by disengaged or intrusive parental stimulation to which the infant responds with flat or negative affect (Field, 1983; Stern, 1985; Tronick & Gianino, 1986). The quality of parent-infant relationship was operationalized through the Parent-Infant Pediatric Examination (PIPE) (Fiese, Poehlmann, Irwin, Gordon, & Curry-Bleggi, 2001).

The psychological impact on parents was operationalized through the assessment of the following variables:

c) *Impact of the infant's CL/P malformation on the family*: This variable is defined as the parental perceptions of the impact of the infant's medical condition on the family (Stein & Reissman, 1980), this is to say, any change in the normative behavior of the family which is directly attributable to the infant's medical condition. Authors argued that changes occur in the family because of illness, forcing adaptations in the family environment. This variable was measure by the French version of the Impact On Family Scale (IOFS) (Boudas et al., 2013).

d) *Psychological distress*: It is defined as a state of emotional suffering characterized by symptoms of depression (e.g., lost interest; sadness; hopelessness) and anxiety (e.g., restlessness; feeling tense) (Mirowsky & Ross 2002), that may impact on the social functioning and day-to-day living of individuals (Wheaton, 2007). Similarly, the concept of psychological distress refers to a general index of psychological alteration grouping together various affective and cognitive symptoms such as depression, anxiety or irritability (Ilfeld, 1976, 1978). This variable was measured with the IDP (*Indice de*

*Détresse Psychologique Enquête Santé Québec-short version) (*Préville, Boyer, Potvin, Perrault, & Légaré, 1992*).*

e) *Postpartum depression*: This variable is defined as moderate to severe depression in a woman after she has given birth. It may occur soon after delivery or up to a year later. DSM-5 (American Psychiatric Association, 2013) does not recognize postpartum depression as a separate diagnosis; rather, patients must meet the criteria for a major depressive episode and the criteria for the peripartum-onset specifier. For research purposes, postpartum depression includes depressions occurring in the first year of life of the child (Gressier, Tabat-Bouher, Cazas, & Hardy, 2015). Depression also affects men during the postnatal period and in later child-rearing years (Goodman, 2004). In the current research, depression symptoms in the course of the first year of the infant was measured thought the French version of the Edinburgh Postpartum Depression Scale (EPDS) (Guedeney & Fermanian, 1998). Although the EPDS was developed to screen for depression in women postnatally, it has been shown to be useful in the assessment of mothers outside the postnatal period (Cox, Chapman, Murray, & Jones, 1996; Murray & Carothers, 1990) and has been validated in men (Matthey et al., 2001).

f) *Marital satisfaction*: This variable is defined as "a subjective experiencing of one's own personal happiness and contentment in the marital relationship" (Hendrick & Hendrick, 1997; p. 57). Marital adjustment -as it is also named- is conceived as a process based on a high degree of agreement between the couple, a low frequency of conflicts and negative interaction, a high frequency of activities in common, and few affective or sexual problems (Spanier, 1976). Marital satisfaction was evaluated by the French version of the Dyadic Adjustment Scale (DAS) (Antoine, Christophe, & Nandrino, 2008), which assess the marital adjustment over the preceding 12 months.

2.6.2. Methodology: non-experimental.

2.6.3. Study design: *prospective, descriptive, longitudinal, within-subjects, and between-groups.*

2.6.4. Data analysis

First, a descriptive analysis of the items conforming the evaluation instruments was performed. The mean, standard deviation, median, mode, asymmetry and kurtosis were provided. Secondly, a descriptive analysis of the total scores for each instrument was performed. The mean, standard deviation, median, mode, asymmetry and kurtosis was provided, minimum and maximum score, and percentiles were obtained.

Thirdly, it was performed an analysis of variance (ANOVA within subjects) for repeated measures in order to know the evolution of the dependent variables through the first (T0) and the second evaluation (T1). Finally, it was carried out an inter-subject analysis of variance (ANOVA between groups) for each independent variable. Type of malformation was separated into two groups: Cleft Lip group and Cleft Lip and Palate group; laterality of the malformation was divided into: unilateral and bilateral; type of diagnosis was grouped in prenatal diagnosis group and diagnosis at birth group; and waiting time prior to the first surgery was divided into: early surgery group (0-90 waiting days prior to surgery), and late surgery group (more than 90 waiting days prior to surgery). All the analyses were performed with the SPSS statistical software, with 95% confidence intervals.

Regarding the interpretation of skewness and kurtosis, these are measures that describes the shape of the data's distribution, this is to say, to identify the normality of the data. There are several appraisals about the interpretation of the values that report a severe deviation from the normally data distribution. Some authors point out that skewness and kurtosis values between -2 and +2 are considered

acceptable in order to approve the normal univariate distribution (George & Malley, 2010; Tabachnich & Fidell, 2013). Other authors maintain the value +/-2 for skewness, but indicate a value of +/-7 for kurtosis (Hair, Black, Babin, & Andersen, 2010).

Moreover, Kline (2011) suggests that the deviation from the normality data is not severe if the values of skewness and kurtosis are between 3 and 10 (absolute values) respectively. For analysis data in the current research, this last recommendation was taken into account. Thus, if skewness and kurtosis values are within the range recommended by Kline (2011), then the data used in the study can be considered to fit a normal distribution.

Finally, with the aim to measure the size of the effect, it has been used the proportion of variance explained (η^2) and its interpretation was based on the criterion of Cohen (1988), in which η^2 =.01 is a small effect; η^2 =.06 is a median effect; and η^2 =.14 is a big effect (Frías-Navarro, 2011).

3. RESULTS

3.1. Descriptive outcomes of the instruments: item analysis

3.1.1. Infant social withdrawal behavior (ADBB)

First of all, the results of descriptive analysis of the eight items of the ADBB scale are presented below. It is important to consider that each item is scored from 0 to 4, according to the infant's performance in the dimension being assessed. Higher scores reflect greater social withdrawal behavior.

Table 3.1 shows the main descriptive statistics of the ADBB items in the first assessment. As can be seen, item 5 "vocalizations" obtained the highest mean (M = 0.45, SD = 0.67), while item 3 "general level of activity" showed the lowest mean (M = 0.16, SD = 0.39). This indicates that, in general, infants with CL/P had more difficulties in the oral expression (less positive and negative vocalizations), and they performed better at the level of corporal activity. In addition, the mode values were zero in all cases, indicating the lack of signals of social withdrawal in most of infants having a CL/P.

Table 3.1. *Main descriptive statistics of the items constituting ADBB scale in the first evaluation (T0)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Facial expression	0.39	0.63	0	0	1.53	1.95	0-3
2. Eye contact	0.21	0.51	0	0	2.48	5.23	0-2
3. General level of activity	**0.16**	0.39	0	0	2.25	4.21	0-2
4. Self-stimulating gestures	0.17	0.49	0	0	**3.34**	**12.00**	0-3
5. Vocalizations	**0.45**	0.67	0	0	1.35	1.07	0-3
6. Response to stimulation	0.28	0.51	0	0	1.60	1.68	0-2
7. Relationship	0.33	0.58	0	0	1.57	1.46	0-2
8. Attraction	0.39	0.65	0	0	1.57	1.84	0-3

Regarding the distribution of data in the first evaluation, it was observed that most of the items had a normal distribution, with the exception of item 4 "self-stimulating gestures", in which the absolute values of asymmetry and kurtosis exceed 3 and 10, respectively.

As regards the second evaluation of infant social withdrawal behavior, table 3.2 shows the results of the descriptive analysis of the eight items constituting the ADBB. Similar to the first evaluation, item 5 "vocalizations" had the highest mean (*M* = 0.38, *SD* = 0.58), and item 3 "general level of activity" obtained the lowest mean (*M* = 0.07, *SD* = 0.25).

That is, at twelve months, infants with CL/P continued to present fewer vocalizations (both positive and negative), and showed a better performance in the area of body activity. In addition, the values of mode and median for the eight items were zero, which would indicate that, in general, the infants had a good performance in the dimensions that evaluates each item of the ADBB scale.

Table 3.2. *Main descriptive statistics of the items constituting ADBB scale in the second evaluation (T1)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Facial expression	0.29	0.49	0	0	1.34	0.71	0-2
2. Eye contact	0.13	0.38	0	0	**3.07**	9.44	0-2
3. General level of activity	**0.07**	0.25	0	0	**3.57**	**10.93**	0-1
4. Self-stimulating gestures	0.08	0.40	0	0	**5.51**	**32.67**	0-3
5. Vocalizations	**0.38**	0.58	0	0	1.24	0.57	0-2
6. Response to stimulation	0.12	0.33	0	0	2.34	3.53	0-1
7. Relationship	0.21	0.43	0	0	1.74	1.91	0-2
8. Attraction	0.26	0.48	0	0	1.56	1.47	0-2

In relation to the normality of the data, it can be considered that most of the items had a normal distribution, with the exception of items 2, 3 and 4, which obtained absolute values of asymmetry and kurtosis that exceeded the ranges proposed for the normal distribution.

3.1.2. Infant social withdrawal behavior (m-ADBB)

The results of descriptive analysis of the five items of the m-ADBB scale are presented below. It is important to consider that each item is scored from "0=Satisfactory," "1=Possible problem," to "2=Definite problem", according to the infant's performance in each dimension assessed. Scores higher than 2 indicate the presence of social withdrawal.

The main descriptive statistics of the m-ADBB items in the first assessment are detailed in table 3.3. As can be seen, item 4 "vocalizations" obtained the highest mean (M = 0.46, SD = 0.69), while item 3 "general level of activity" showed the lowest mean (M = 0.18, SD = 0.41). This reflects similar findings that those with the ADBB scale: infants with cleft lip and palate (CL/P) had more difficulties in expressing positive and negative vocalizations, and the dimension with less alterations was corporal activity. The results of the analysis also revealed that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.3. *Main descriptive statistics of the items constituting m-ADBB scale in the first evaluation (T0)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Facial expression	0.44	0.64	0	0	1.33	1.39	0-3
2. Eye contact	0.18	0.49	0	0	2.80	7.00	0-2
3. General level of activity	**0.18**	0.41	0	0	1.98	2.90	0-2
4. Vocalizations	**0.46**	0.69	0	0	1.31	0.93	0-3
5. Relationship	0.40	0.60	0	0	1.21	0.44	0-2

In the second assessment of social withdrawal, through the m-ADBB scale, table 3.4 shows the descriptive statistics of the five items. As can be observed, item 4 "vocalizations" had the highest mean (*M* = 0.36, *SD* = 0.56), and item 3 "general level of activity" obtained the lowest mean (*M* = 0.07, *SD* = 0.25).

At twelve months, infants with CL/P still had more alterations in the vocalizations area, while the dimension of corporal activity had also the better performance. Moreover, most of the items were within the ranges of normal distribution, only items 2 and 3 presented absolute values superior to 3 and 10 in asymmetry and kurtosis, respectively.

Table 3.4. *Main descriptive statistics of the items constituting m-ADBB scale in the second evaluation (T1)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Facial expression	0.28	0.49	0	0	1.39	0.88	0-2
2. Eye contact	0.11	0.34	0	0	**3.06**	9.32	0-2
3. General level of activity	**0.07**	0.25	0	0	**3.57**	**10.93**	0-1
4. Vocalizations	**0.36**	0.56	0	0	1.30	0.74	0-2
5. Relationship	0.20	0.40	0	0	1.56	0.43	0-1

3.1.3. Quality of the parent-infant relationship (PIPE)

The results of the descriptive analysis of the three items that constitute the PIPE instrument at four (T0) and twelve months (T1) are presented below. Each item of the PIPE scale evaluates a time of the interaction (starting the game, keeping the game going and stopping the game). Each item can be rated from zero to five. The lower scores suggest a more adaptive or higher quality parent-infant interaction.

In table 3.5 it can be seen that, at four months, item 2 "keeping the game going" obtained the highest mean (*M* = 0.78, *SD* = 1.10), while item 1 "starting the game" had the lower mean (*M* = 0.64, *SD* = 1.06). This would indicate that there were greater alterations in the parent-infant relationship during the game development, and that the higher quality of the interaction was presented at the beginning. Moreover, for the three items, the median and the mode were zero, which would indicate a low frequency of alterations in the parent-baby relationship. In addition, the results of this analysis revealed that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.5. *Main descriptive statistics of the items constituting the PIPE in the first evaluation (T0)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Starting the game	0.64	1.06	0	0	1.97	3.65	0-5
2. Keeping the game going	0.78	1.10	0	0	1.65	2.39	0-5
3. Stopping the game	0.76	1.19	0	0	1.96	3.93	0-5

Regarding the second evaluation, Table 3.6 details the main descriptive of the three items that constitute the PIPE scale. As can be seen, item 2 "keeping the game going" obtained the highest mean (*M* = 0.55, *SD* = 0.81), while item 1 "starting the game" had the lowest mean (*M* = 0.45, *SD* = 0.84). These results would indicate that there were greater difficulties during the development of the parent-infant game, and that the quality of the relationship was greater at the beginning. Besides, the values for the median and the mode were zero in the three items.

That is, the majority of the families did not present alterations in the interaction. Concerning the distribution of the data, the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.6. *Main descriptive statistics of the items constituting the PIPE in the second evaluation (T1)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Starting the game	0.45	0.84	0	0	1.97	3.65	0-5
2. Keeping the game going	0.55	0.81	0	0	1.65	2.39	0-4
3. Stopping the game	0.51	0.90	0	0	1.96	3.93	0-5

3.1.4. Impact on family (IOFS)

3.1.4.1. Impact on family reported by mothers

The results of the descriptive analysis of the fifteen items that constitute the IOFS instrument, evaluated in mothers at four months (T0), are detailed in table 3.7. It should be noted that each of the items on the IOFS scale can be scored from one to four, depending on the degree of agreement with the statement. Higher scores suggest a greater impact of the malformation in the family.

As can be seen in table 3.7, item 14 "Travel to hospital is a strain" had the highest mean (*M* = 2.09, *SD* = 0.92), this is to say, the greatest impact of malformation for mothers resided in travel to the hospital. On the contrary, item 6 "See family and friends less" obtained the lowest mean (*M* = 1.26, *SD* = 0.54), this would mean that the mothers reported a lower impact of their infant's malformation in relation to their social life.

Mode and median values ranged from one to two, which would indicate that the impact on the family was generally moderate to low. Regarding the distribution of the data, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.38 to 2.26, kurtosis: 0.00 to 5.58).

Table 3.7. *Main descriptive statistics of the items constituting the IOFS in the first evaluation (T0) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Can't travel out of city	1.31	0.58	1	1	1.74	1.95	1-3
2. People treat us special	1.47	0.68	1	1	1.28	0.89	1-4
3. Little desire to go out	1.38	0.71	1	1	2.04	3.88	1-4
4. Hard to find reliable person to care for child	1.89	0.95	2	1	0.79	-0.38	1-4
5. Need to change plans at last minute	1.52	0.74	1	1	1.24	0.73	1-4
6. See family and friends less	**1.26**	0.54	1	1	2.26	5.58	1-4
7. Wonder whether to treat child "specially"	1.71	0.85	1	1	0.96	-0.00	1-4
8. Think about not having more children	1.68	0.99	1	1	1.27	0.36	1-4
9. No time for other family members	1.56	0.72	1	1	1.25	1.34	1-4
10. Family gives up things	1.39	0.61	1	1	1.30	0.63	1-3
11. Fatigue is a problem	1.49	0.71	1	1	1.36	1.24	1-4
12. Live from day to day	1.50	0.79	1	1	1.58	1.87	1-4
13. Nobody understands the burden	1.73	0.78	2	1	0.89	0.38	1-4
14. Travel to hospital is a strain	**2.09**	0.92	2	2	0.38	-0.77	1-4
15. Live on roller coaster	2.02	0.95	2	1	0.42	-0.93	1-4

Table 3.8 shows the results of the descriptive analysis of the fifteen items that constitute the IOFS instrument evaluated in mothers at twelve months. Similar to the first evaluation, item 14 "Travel to hospital is a strain" had the highest mean (*M* = 1.89, *SD* = 0.90). Therefore, at 12 months, the major impact of the malformation, according to the mothers, remained to be the displacement to the hospital. In contrast, the lowest means were observed in items 1 "Cannot travel out of city" and 3 "Little desire to go out".

Mode and median values ranged from one to two, suggesting a low-modest impact of the malformation on the family. In addition, the results showed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.63 to 2.82, kurtosis: 0.23 to 7.66).

Table 3.8. *Main descriptive statistics of the items constituting the IOFS in the second evaluation (T1) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Can't travel out of city	**1.16**	0.43	1	1	2.82	7.66	1-3
2. People treat us special	1.25	0.49	1	1	1.82	2.54	1-3
3. Little desire to go out	**1.16**	0.39	1	1	2.34	4.77	1-3
4. Hard to find reliable person to care for child	1.69	0.84	1	1	0.89	-0.23	1-4
5. Need to change plans at last minute	1.40	0.69	1	1	1.62	1.74	1-4
6. See family and friends less	1.17	0.40	1	1	2.13	3.69	1-3
7. Wonder whether to treat child "specially"	1.49	0.83	1	1	1.74	2.28	1-4
8. Think about not having more children	1.60	0.94	1	1	1.43	0.93	1-4
9. No time for other family members	1.37	0.61	1	1	1.64	2.68	1-4
10. Family gives up things	1.17	0.40	1	1	2.23	4.20	1-3
11. Fatigue is a problem	1.24	0.52	1	1	2.46	7.27	1-4
12. Live from day to day	1.42	0.67	1	1	1.34	0.51	1-3
13. Nobody understands the burden	1.58	0.77	1	1	1.11	0.35	1-4
14. Travel to hospital is a strain	**1.89**	0.90	2	2	0.63	-0.59	1-4
15. Live on roller coaster	1.83	0.94	2	1	0.82	-0.39	1-4

3.1.4.2. Impact on family reported by fathers

Concerning the fathers, the results of the descriptive analysis of the fifteen items of the IOFS instrument, evaluated at four months (T0), are detailed in table 3.9. Similar to mothers, item 14 "Travel to hospital is a strain" had the highest mean (*M* = 2.16, *SD* = 0.91). That is to say, the major impact of the malformation was the displacement to hospital. The lowest mean was observed in item 6 "See family and friends less", which would indicate less impact of the malformation of their infant in the social aspect of the fathers.

Mode and median ranged from one to two, indicating a low-modest impact of the malformation on the family. Also, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.26 to 2.29, kurtosis: 0.39 to 5.97).

Table 3.9. *Main descriptive statistics of the items constituting the IOFS in the first evaluation (T0) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Can't travel out of city	1.36	0.64	1	1	1.56	1.25	1-3
2. People treat us special	1.54	0.68	1	1	1.03	0.52	1-4
3. Little desire to go out	1.40	0.70	1	1	1.77	2.61	1-4
4. Hard to find reliable person to care for child	1.87	0.95	2	1	0.73	-0.56	1-4
5. Need to change plans at last minute	1.48	0.70	1	1	1.57	2.52	1-4
6. See family and friends less	**1.26**	0.54	1	1	2.29	5.97	1-4
7. Wonder whether to treat child "specially"	1.63	0.87	1	1	1.40	1.31	1-4
8. Think about not having more children	1.44	0.75	1	1	1.70	2.15	1-4
9. No time for other family members	1.60	0.71	1	1	1.02	0.79	1-4
10. Family gives up things	1.38	0.57	1	1	1.16	0.39	1-3
11. Fatigue is a problem	1.45	0.60	1	1	1.42	3.18	1-4
12. Live from day to day	1.50	0.79	1	1	1.55	1.71	1-4
13. Nobody understands the burden	1.60	0.72	1	1	1.05	0.70	1-4
14. Travel to hospital is a strain	**2.16**	0.91	2	2	0.26	-0.82	1-4
15. Live on roller coaster	1.94	0.94	2	1	0.58	-0.75	1-4

Table 3.10 shows the results of the descriptive analysis of the fifteen items that form the IOFS instrument evaluated in the fathers at twelve months (T1). As can be seen, item 14 "Travel to hospital is a strain" had the highest mean (*M* = 1.82, *SD* = 0.82). This confirms that in all cases, the major impact of the malformation was on journeys to the hospital. On the contrary, the lowest means were observed in items 1 "Cannot travel out of city", 3 "Little desire to go out", 5 "Need to change plans at last minute" and 10 "Family gives up things".

Median values ranged from one to two, while mode was one for all items. This would indicate a low-moderate impact on the family, according to fathers. Regarding the distribution of the data, the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.82 to 2.37, kurtosis: 0.17 to 5.06).

Table 3.10. *Main descriptive statistics of the items constituting the IOFS in the second evaluation (T1) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Can't travel out of city	**1.16**	0.40	1	1	2.37	5.06	1-3
2. People treat us special	1.23	0.47	1	1	1.89	2.83	1-3
3. Little desire to go out	**1.16**	0.37	1	1	1.88	1.58	1-2
4. Hard to find reliable person to care for child	1.62	0.86	1	1	1.31	0.91	1-4
5. Need to change plans at last minute	**1.16**	0.40	1	1	2.37	5.06	1-3
6. See family and friends less	1.17	0.40	1	1	2.24	4.37	1-3
7. Wonder whether to treat child "specially"	1.51	0.75	1	1	1.38	1.32	1-4
8. Think about not having more children	1.44	0.81	1	1	1.72	1.89	1-4
9. No time for other family members	1.34	0.54	1	1	1.28	0.68	1-3
10. Family gives up things	**1.16**	0.37	1	1	1.88	1.58	1-2
11. Fatigue is a problem	1.24	0.43	1	1	1.24	-0.48	1-2
12. Live from day to day	1.38	0.65	1	1	1.71	2.59	1-4
13. Nobody understands the burden	1.52	0.72	1	1	1.35	1.58	1-4
14. Travel to hospital is a strain	**1.82**	0.82	2	1	0.82	-0.17	1-4
15. Live on roller coaster	1.68	0.82	1	1	1.00	0.26	1-4

3.1.5. Psychological distress (IDP)

3.1.5.1. Psychological distress reported by mothers

The results of the descriptive analysis of the fourteen items that form the IDP instrument are presented below. Each of the items can be scored with one or two, depending on the degree of agreement with the statement. Higher scores indicate greater psychological distress. As can be seen in table 3.11, in the first evaluation, item 2 "Did you feel fearful or afraid?" obtained the highest mean (*M* = 1.63, *SD* = 0.48). This would mean that mothers tended to have high rates of fear related to their infant's malformation. In contrast, the lowest means were presented in items 4 and 7, which refer to depression, specifically to feel hopeless about the future and to have little interest in things.

In addition, the results of the analysis showed that the absolute values of asymmetry and kurtosis were within normal distribution ranges (asymmetry: 0.06 to 2.84, kurtosis: 0.02 to 6.13).

Table 3.11. *Main descriptive statistics of the items constituting the IDP in the first evaluation (T0) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Did you feel tense or under pressure?	1.53	0.50	2	2	-0.12	-0.02	1-2
2. Did you feel fearful or afraid?	**1.63**	0.48	2	2	-0.56	-1.71	1-2
3. Did you feel nervous or shaky inside?	1.56	0.50	2	2	-0.24	-1.97	1-2
4. Did you feel hopeless about the future?	**1.09**	0.29	1	1	2.84	6.13	1-2
5. Did you feel lonely?	1.20	0.40	1	1	1.52	0.30	1-2
6. Did you feel downhearted or blue?	1.38	0.49	1	1	0.51	-1.77	1-2
7. Did you feel bored or have little interest in things?	**1.09**	0.29	1	1	2.82	6.05	1-2
8. Did you cry easily or feel like crying?	1.61	0.49	2	2	-0.44	-1.83	1-2
9. Did you lose your temper?	1.46	0.50	1	1	0.15	-2.01	1-2
10. Did you feel critical of others?	1.14	0.35	1	1	2.06	2.29	1-2
11. Did you feel easily annoyed or irritated?	1.51	0.50	2	2	-0.06	-2.03	1-2
12. Did you get angry over things that are not too important?	1.47	0.50	1	1	0.10	-2.02	1-2
13. Did you have your mind go blank?	1.19	0.39	1	1	1.63	0.68	1-2
14. Did you have trouble remembering things?	1.22	0.42	1	1	1.36	-0.16	1-2

Table 3.12 details the results of the descriptive analysis of the fourteen items that form the IDP instrument evaluated in the mothers at twelve months (T1). As can be seen, item 11 "Did you feel easily annoyed or irritated?" had the highest mean (*M* = 1.54, *SD* = 0.50). That is, at twelve months, mothers often had high rates of irritability. On the contrary, item 4 -which refers to feeling hopeless before the future- obtained the lowest mean (*M* = 1.07, *SD* = 0.26).

In addition, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.00 to 3.30, kurtosis: 0.19 to 9.05), except for item 4 whose asymmetry value exceeded the absolute value of three.

Table 3.12. *Main descriptive statistics of the items constituting the IDP in the second evaluation (T1) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Did you feel tense or under pressure?	1.51	0.50	2.00	2	-0.03	-2.03	1-2
2. Did you feel fearful or afraid?	1.50	0.50	1.50	1ª	0.00	-2.03	1-2
3. Did you feel nervous or shaky inside?	1.50	0.50	1.50	1ª	0.00	-2.03	1-2
4. Did you feel hopeless about the future?	**1.07**	0.26	1.00	1	**3.30**	9.05	1-2
5. Did you feel lonely?	1.24	0.43	1.00	1	1.25	-0.45	1-2
6. Did you feel downhearted or blue?	1.39	0.49	1.00	1	0.44	-1.84	1-2
7. Did you feel bored or have little interest in things?	1.17	0.38	1.00	1	1.76	1.11	1-2
8. Did you cry easily or feel like crying?	1.45	0.50	1.00	1	0.19	-2.00	1-2
9. Did you lose your temper?	1.47	0.50	1.00	1	0.13	-2.02	1-2
10. Did you feel critical of others?	1.20	0.40	1.00	1	1.48	0.19	1-2
11. Did you feel easily annoyed or irritated?	**1.54**	0.50	2.00	2	-0.17	-2.01	1-2
12. Did you get angry over things that are not too important?	1.52	0.50	2.00	2	-0.08	-2.03	1-2
13. Did you have your mind go blank?	1.17	0.38	1.00	1	1.76	1.11	1-2
14. Did you have trouble remembering things?	1.22	0.42	1.00	1	1.35	-0.19	1-2

3.1.5.2. Psychological distress reported by fathers

Regarding the fathers' evaluation, table 3.13 shows the results of the descriptive analysis of the fourteen items of the IDP, evaluated at four months (T0). As can be seen, item 2 "Did you feel fearful or afraid?" got the highest average (*M* = 1.51, *SD* = 0.50). In other words, fathers were often afraid. On the contrary, item 5 –which refers to feeling alone- showed the lowest mean (*M* = 1.08, *SD* = 0.27). This is to say, the feeling of loneliness was rare.

Moreover, it was also observed that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution (asymmetry: 0.05 to 3.15, kurtosis: 0.12 to 8.05), except for item 5, whose asymmetry value exceeded the absolute value of three.

Table 3.13. *Main descriptive statistics of the items constituting the IDP in the first evaluation (T0) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Did you feel tense or under pressure?	1.44	0.50	1	1	0.25	-1.97	1-2
2. Did you feel fearful or afraid?	**1.51**	0.50	2	2	-0.05	-2.03	1-2
3. Did you feel nervous or shaky inside?	1.46	0.50	1	1	0.18	-2.00	1-2
4. Did you feel hopeless about the future?	1.10	0.31	1	1	2.64	5.05	1-2
5. Did you feel lonely?	**1.08**	0.27	1	1	**3.15**	8.05	1-2
6. Did you feel downhearted or blue?	1.21	0.41	1	1	1.46	0.12	1-2
7. Did you feel bored or have little interest in things?	1.16	0.37	1	1	1.89	1.60	1-2
8. Did you cry easily or feel like crying?	1.25	0.44	1	1	1.14	-0.70	1-2
9. Did you lose your temper?	1.35	0.48	1	1	0.64	-1.62	1-2
10. Did you feel critical of others?	1.20	0.40	1	1	1.53	0.49	1-2
11. Did you feel easily annoyed or irritated?	1.40	0.49	1	1	0.39	-1.88	1-2
12. Did you get angry over things that are not too important?	1.32	0.47	1	1	0.80	-1.39	1-2
13. Did you have your mind go blank?	1.17	0.38	1	1	1.74	1.03	1-2
14. Did you have trouble remembering things?	1.20	0.40	1	1	1.53	0.35	1-2

In the second evaluation, table 3.14 shows the results for fathers of the descriptive analysis of the fourteen items of the IDP. As can be noted, item 12 "Did you get angry over things that are not too important?" obtained the highest mean (*M* = 1.39, *SD* = 0.49), suggesting a high presence of irritability in fathers at twelve months. On the contrary, item 5, referring to the feeling of loneliness, obtained the lowest mean (*M* = 1.04, *SD* = 0.20).

The values of median and mode was one in all cases. Regarding the normality of the data, most of the items showed a normal distribution, except for items 4 and 5 which surpassed the absolute values of 3 and 10 for asymmetry and kurtosis, respectively.

Table 3.14. *Main descriptive statistics of the items constituting the IDP in the second evaluation (T1) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. Did you feel tense or under pressure?	1.35	0.48	1	1	0.26	-1.64	1-2
2. Did you feel fearful or afraid?	1.31	0.47	1	1	0.82	-1.36	1-2
3. Did you feel nervous or shaky inside?	1.38	0.49	1	1	0.49	-1.79	1-2
4. Did you feel hopeless about the future?	1.07	0.25	1	1	**3.46**	**10.20**	1-2
5. Did you feel lonely?	**1.04**	0.20	1	1	**4.82**	**21.65**	1-2
6. Did you feel downhearted or blue?	1.16	0.37	1	1	1.92	1.70	1-2
7. Did you feel bored or have little interest in things?	1.13	0.34	1	1	2.27	3.21	1-2
8. Did you cry easily or feel like crying?	1.16	0.37	1	1	1.92	1.70	1-2
9. Did you lose your temper?	1.36	0.48	1	1	0.58	-1.70	1-2
10. Did you feel critical of others?	1.15	0.36	1	1	2.02	2.13	1-2
11. Did you feel easily annoyed or irritated?	1.37	0.49	1	1	0.36	-1.75	1-2
12. Did you get angry over things that are not too important?	**1.39**	0.49	1	1	0.45	-1.84	1-2
13. Did you have your mind go blank?	1.12	0.32	1	1	2.41	3.88	1-2
14. Did you have trouble remembering things?	1.20	0.40	1	1	1.55	0.42	1-2

3.1.6. Postnatal depression (EPDS)

3.1.6.1. Postnatal depression reported by mothers

The results of descriptive analysis of the ten items that constitute the EPDS are detailed below. It should be noted that each item can receive a score of zero to three, depending on the option that best describes the emotional state of the mother or the father. Higher scores indicate greater depressive symptoms.

Table 3.15 shows that, in the first evaluation, item 3 "I have blamed myself unnecessarily when things went wrong" obtained the highest mean *(M* = 1.41, *SD* = 0.87). That is, at four months, mothers frequently had feelings of guilt when things did not go well. On the contrary, item 10, which refers to self-harm, had the lowest mean (*M* = 0.14, *SD* = 0.50). That is, this type of thinking was the least frequent in mothers. In relation to the normality of the data, the majority of the items had a normal distribution, except for the item 10, in which the asymmetry and kurtosis values exceeded the absolute values of three and ten, respectively.

Table 3.15. *Main descriptive statistics of the items constituting the EPDS in the first evaluation (T0) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. I have been able to laugh and see the funny side of things	0.35	0.59	0.00	0	1.45	1.08	0-2
2. I have looked forward with enjoyment to things	0.33	0.60	0.00	0	1.89	3.30	0-3
3. I have blamed myself unnecessarily when things went wrong	**1.41**	0.87	1.50	2	-0.13	-0.74	0-3
4. I have been anxious or worried for no good reason	1.24	0.95	1.00	2	-0.04	-1.18	0-3
5. I have felt scared or panicky for no very good reason	0.82	0.87	1.00	0	-0.68	-0.56	0-3
6. Things have been getting on top of me	1.09	0.70	1.00	1	0.13	-0.34	0-3
7. I have been so unhappy that I have had difficulty sleeping	0.55	0.82	0.00	0	1.34	0.83	0-3
8. I have felt sad or miserable	0.67	0.74	1.00	0	1.08	1.24	0-3
9. I have been so unhappy that I have been crying	0.56	0.79	0.00	0	1.44	1.58	0-3
10. The thought of harming myself has occurred to me	**0.14**	0.50	0.00	0	**3.74**	**14.08**	0-3

Regarding the second evaluation of mothers, table 3.16 details the results of the descriptive analysis of the ten items of the EPDS. Similar to the first evaluation, item 3 "I have blamed myself unnecessarily when things went wrong" had the highest mean (*M* = 1.46, *SD* = 0.87), and item 10 -referring to self-harm- obtained the lowest mean (*M* = 0.13, *SD* = 0.44). The analysis showed that most of the items had a normal distribution, with the exception of item 10, in which the values of asymmetry and kurtosis exceeded the absolute values of three and ten, respectively.

Table 3.16. *Main descriptive statistics of the items constituting the EPDS in the second evaluation (T1) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. I have been able to laugh and see the funny side of things	0.38	0.68	0	0	1.84	3.00	0-3
2. I have looked forward with enjoyment to things	0.31	0.71	0	0	2.48	5.62	0-3
3. I have blamed myself unnecessarily when things went wrong	**1.46**	0.87	2	2	-0.35	-0.73	0-3
4. I have been anxious or worried for no good reason	1.15	0.92	1	2	-0.05	-1.34	0-3
5. I have felt scared or panicky for no very good reason	0.67	0.78	0	0	-0.77	-0.56	0-3
6. Things have been getting on top of me	0.98	0.75	1	1	0.39	-0.16	0-3
7. I have been so unhappy that I have had difficulty sleeping	0.54	0.79	0	0	1.21	0.36	0-3
8. I have felt sad or miserable	0.73	0.81	1	0	1.02	0.63	0-3
9. I have been so unhappy that I have been crying	0.50	0.68	0	0	1.52	2.92	0-3
10. The thought of harming myself has occurred to me	**0.13**	0.44	0	0	**3.57**	**11.81**	0-2

3.1.6.2. Postnatal depression reported by fathers

Table 3.17 shows the results of the descriptive analysis of the ten items of the EPDS, in the first evaluation of fathers. Similar to mothers, item 3 "I have blamed myself unnecessarily when things went wrong" had the highest mean (*M* = 1.09, *SD* = 0.97) and item 10 -referring to self-harm- obtained the lowest mean (*M* = 0.14, *SD* = 0.49). That is, in fathers, the feeling of guilt and self-reproach was the most frequent, while thoughts related to harm were the least recurrent.

Concerning the normality of the data, most of the items had a normal distribution, with the exception of item 10, in which the asymmetry and kurtosis exceeded the absolute values of three and ten, respectively.

Table 3.17. *Main descriptive statistics of the items constituting the EPDS in the first evaluation (T0) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. I have been able to laugh and see the funny side of things	0.33	0.60	0	0	1.89	3.44	0-3
2. I have looked forward with enjoyment to things	0.28	0.60	0	0	2.45	6.46	0-3
3. I have blamed myself unnecessarily when things went wrong	**1.09**	0.97	1	0	0.34	-1.01	0-3
4. I have been anxious or worried for no good reason	0.92	0.96	1	0	0.48	-1.11	0-3
5. I have felt scared or panicky for no very good reason	0.61	0.85	0	0	1.08	-0.04	0-3
6. Things have been getting on top of me	0.87	0.70	1	1	0.61	0.59	0-3
7. I have been so unhappy that I have had difficulty sleeping	0.52	0.79	0	0	1.36	0.92	0-3
8. I have felt sad or miserable	0.52	0.61	0	0	0.97	1.13	0-3
9. I have been so unhappy that I have been crying	0.15	0.50	0	0	4.05	18.27	0-3
10. The thought of harming myself has occurred to me	**0.14**	0.49	0	0	**3.93**	**15.76**	0-3

The results of the descriptive analysis of the ten items of the EPDS are presented below, in the second evaluation of fathers (T1). Table 3.18 shows that item 3 "I have blamed myself unnecessarily when things went wrong" obtained the highest mean (*M* = 0.98, *SD* = 0.55), and item 10 -referring to self-harm - had the lowest mean (*M* = 0.10, *SD* = 0.42). In other words, fathers showed greater feelings of guilt and less thoughts about self-harm.

In relation to the normality of the data, most of the items had a normal distribution, with the exception of item 10, in which the asymmetry and kurtosis exceeded the absolute values of three and ten, respectively.

Table 3.18. *Main descriptive statistics of the items constituting the EPDS in the second evaluation (T1) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1. I have been able to laugh and see the funny side of things	0.18	0.46	0	0	2.60	6.29	0-2
2. I have looked forward with enjoyment to things	0.19	0.55	0	0	**3.54**	**13.99**	0-3
3. I have blamed myself unnecessarily when things went wrong	**0.98**	0.92	1	0	0.36	-1.09	0-3
4. I have been anxious or worried for no good reason	0.72	0.87	0	0	0.77	-0.71	0-3
5. I have felt scared or panicky for no very good reason	0.45	0.76	0	0	1.46	-0.88	0-3
6. Things have been getting on top of me	0.76	0.75	1	0	0.58	-0.53	0-3
7. I have been so unhappy that I have had difficulty sleeping	0.33	0.62	0	0	1.97	3.76	0-3
8. I have felt sad or miserable	0.48	0.74	0	0	1.84	3.55	0-3
9. I have been so unhappy that I have been crying	0.12	0.32	0	0	2.42	3.95	0-1
10. The thought of harming myself has occurred to me	**0.10**	0.42	0	0	**4.30**	**17.19**	0-2

3.1.7. Marital satisfaction (DAS)

3.1.7.1. Marital satisfaction reported by mothers

This section details the results of the descriptive analysis of the sixteen items constitute the DAS. It should be mentioned that each of the items can be scored from one to six, depending on the degree of agreement with the statements. Higher scores indicate greater marital satisfaction.

As can be seen in table 3.19, in the first evaluation, item 8 "Divorce" had the highest mean (*M* = 5.38, *SD* = 0.95). This would indicate that divorce was the least frequent thought in mothers at T0. On the contrary, the lowest mean was obtained in item 15, referring to working on a joint project (*M* = 3.85, *SD* = 1.55). Regarding the normality of the data, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.06 to 1.93, kurtosis: 0.25 to 5.13).

Table 3.19. *Main descriptive statistics of the items constituting the DAS in the first evaluation (T0) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1 Objectives	4.78	0.96	5	5	-0.86	0.67	2-6
2 Decisions	5.04	0.96	5	5	-1.50	3.55	1-6
3 Affection	4.89	1.09	5	5	-1.16	1.17	1-6
4 Friends	4.76	1.10	5	5	-0.87	0.60	1-6
5 Sexuality	4.48	1.18	5	5	-0.98	0.79	1-6
6 Life Philosophy	4.47	1.16	5	5	-0.82	0.45	1-6
7 Parents –in law	4.39	1.23	5	5	-0.82	0.29	1-6
8 Divorce	**5.38**	0.95	6	6	-1.93	5.13	1-6
9 Nerves	4.54	0.92	4	4	-0.06	-0.26	2-6
10 Exchange of ideas	4.03	1.03	4	4	-0.19	-0.26	1-6
11 Discussions	4.45	1.09	5	5	-0.77	0.25	1-6
12 Confidence	4.82	1.24	5	6	-1.11	0.65	1-6
13 Common interest	4.19	1.33	4	5	-0.57	-0.35	1-6
14 Laugh together	4.96	0.91	5	5	-0.45	-0.35	2-6
15 Common project	**3.85**	1.55	4	3[a]	-0.24	-0.96	1-6
16 Happiness	4.76	0.93	5	5	-1.09	2.67	1-6

Table 3.20 shows the results of the descriptive analysis of the sixteen items of the DAS in the second evaluation of mothers. Similar to the first assessment, item 8 "Divorce" obtained the highest mean (*M* = 5.32, *SD* = 1.01). This would indicate that divorce was the least frequent thought among mothers. In contrast, the lowest mean was observed in item 15 "Common project" (*M* = 3.89, *SD* = 1.55). Regarding the normality of the data, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.31 to 1.94, kurtosis: 0.07 to 4.30).

Table 3.20. *Main descriptive statistics of the items constituting the DAS in the second evaluation (T1) (mothers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1 Objectives	4.64	0.95	5	5	-0.74	0.32	2-6
2 Decisions	4.95	0.87	5	5	-0.80	0.66	2-6
3 Affection	4.70	1.14	5	5	-0.93	0.60	1-6
4 Friends	4.71	0.98	5	5	-0.66	0.17	2-6
5 Sexuality	4.22	1.22	4	5	-0.46	-0.43	1-6
6 Life Philosophy	4.52	1.00	5	5	-0.51	-0.13	2-6
7 Parents –in law	4.46	1.31	5	5	-0.98	0.51	1-6
8 Divorce	**5.32**	1.01	6	6	-1.94	4.30	1-6
9 Nerves	4.46	1.02	4	4	-0.36	0.53	1-6
10 Exchange of ideas	4.01	1.08	4	4	-0.49	-0.07	1-6
11 Discussions	4.50	1.09	5	5	-0.73	0.29	1-6
12 Confidence	4.71	1.35	6	6	-1.03	0.40	1-6
13 Common interest	3.96	1.35	4	4	-0.47	-0.35	1-6
14 Laugh together	4.70	0.95	5	5	-0.31	-0.50	2-6
15 Common project	**3.89**	1.55	5	5	-0.34	-0.89	1-6
16 Happiness	4.60	1.03	5	5	-1.01	1.84	1-6

3.1.7.2. Marital satisfaction reported by fathers

The results of the descriptive analysis of the sixteen items of the DAS in the first evaluation of the fathers are detailed in table 3.21. Similar to the mothers, item 8 "Divorce" obtained the highest mean (*M* = 5.54, *SD* = 0.88). This would indicate that fathers, like mothers, did not think about divorce or separation. On the contrary, the lowest mean was obtained in item 10 "Exchange of ideas" (*M* = 3.82, *SD* = 1.07). Regarding the normality of the data, the results revealed that the absolute values of asymmetry and kurtosis were within normal distribution ranges (asymmetry: 0.26 to 2.43, kurtosis: 0.33 to 7.07).

Table 3.21. *Main descriptive statistics of the items constituting the DAS in the first evaluation (T0) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1 Objectives	4.76	0.93	5	5	-0.83	0.38	2-6
2 Decisions	5.06	0.83	5	5	-0.78	0.73	2-6
3 Affection	4.94	1.00	5	5	-0.99	1.19	1-6
4 Friends	4.67	1.07	5	5	-0.86	0.60	1-6
5 Sexuality	4.57	1.08	5	5	-0.38	-0.75	2-6
6 Life Philosophy	4.64	1.09	5	5	-1.01	1.31	1-6
7 Parents –in law	4.53	1.14	5	5	-0.50	-0.54	2-6
8 Divorce	**5.54**	0.88	6	6	-2.43	7.07	1-6
9 Nerves	4.66	1.00	5	4	-0.40	0.35	1-6
10 Exchange of ideas	**3.82**	1.07	4	4	-0.50	0.70	1-6
11 Discussions	4.60	1.04	5	5	-0.52	-0.46	2-6
12 Confidence	4.55	1.39	5	6	-0.75	-0.43	1-6
13 Common interest	4.13	1.17	4	5	-0.28	-0.46	1-6
14 Laugh together	4.92	0.96	5	5[a]	-0.44	-0.84	3-6
15 Common project	4.06	1.55	4	3[a]	-0.31	-1.02	1-6
16 Happiness	4.86	0.78	5	5	-0.26	-0.33	3-6

Concerning the second evaluation of the fathers, table 3.22 details the results of the descriptive analysis of the sixteen items that constitute the DAS. Similar to the first evaluation, item 8 "Divorce" obtained the highest mean (*M* = 5.55, *SD* = 0.81). This indicates that divorce was not a recurrent thought in the fathers. On the contrary, the lowest mean was presented in item 10, referring to the exchange of stimulating ideas between the couple (*M* = 3.89, *SD* = 1.05). Regarding the normality of the data, the results revealed that the absolute values of asymmetry and kurtosis were within the normal distribution ranges (asymmetry: 0.12 to 2.61, kurtosis: 0.01 to 9.75).

Table 3.22. *Main descriptive statistics of the items constituting the DAS in the second evaluation (T1) (fathers)*

Items	*Mean*	*SD*	*Median*	*Mode*	*Asymmetry*	*Kurtosis*	*Range*
1 Objectives	4.89	0.82	5	5	-1.04	1.57	2-6
2 Decisions	5.06	0.90	5	5	-1.57	4.35	1-6
3 Affection	4.87	1.09	5	5	-0.96	0.91	1-6
4 Friends	4.75	0.98	5	5	-1.05	1.83	1-6
5 Sexuality	4.37	1.18	5	5	-0.62	-0.23	1-6
6 Life Philosophy	4.58	1.12	5	5	-0.62	-0.02	1-6
7 Parents –in law	4.54	1.12	5	5	-0.62	0.05	1-6
8 Divorce	**5.55**	0.81	6	6	-2.61	9.75	1-6
9 Nerves	4.63	1.05	4	4	-0.67	1.51	1-6
10 Exchange of ideas	**3.89**	1.05	4	4	-0.53	0.38	1-6
11 Discussions	4.53	1.10	5	5	-0.70	-0.18	2-6
12 Confidence	4.44	1.30	5	5	-0.48	-0.71	1-6
13 Common interest	3.93	1.13	4	3	-0.21	-0.40	1-6
14 Laugh together	4.67	1.00	5	5	-0.54	0.01	2-6
15 Common project	4.00	1.39	4	3[a]	-0.12	-1.02	1-6
16 Happiness	4.77	0.83	5	5	-0.74	1.30	2-6

3.2. Descriptive outcomes of the instruments: analysis of the total scores

3.2.1 Infant social withdrawal behavior (ADBB)

The results of the descriptive analysis of the total scores of the ADBB scale are detailed in table 3.21. The theoretical amplitude of the response scale is 0 to 32, with a cut-off score of five. That is, total scores equal or above five indicate the presence of infant social withdrawal behavior. As can be seen, the means of the ADBB total scores were low, in both evaluations. It means that the level of social withdrawal behavior was reduced in this sample of infants with CL/P, at T0 and T1. In addition, the mean was higher in the first evaluation (*M* = 2.38, *SD* = 2.97) compared to the second measurement (*M* = 1.54, *SD* = 2.01).

Regarding the amplitude of the total scores was greater in the first evaluation (T0), compared to the second one (T1). In relation to the distribution of the data, the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.21. Main descriptive statistics of the ADBB scale in the first (T0) and the second (T1) evaluation

		ADBB T0 Total score	ADBB T1 Total score
N	Valid	145	123
	Missing	0	22
Mean		**2.38**	**1.54**
Median		2.00	1.00
Mode		0	0
Standard deviation		2.97	2.01
Asymmetry		2.00	1.92
Std. Error Asymmetry		0.201	0.218
Kurtosis		4.89	5.35
Std. Error Kurtosis		0.400	0.433
Minimum		0	0
Maximum		17	12
Percentile	Percentile 25	0	0
	Percentile 50	2	1
	Percentile 75	3	2

Table 3.22 shows the frequencies of the ADBB total scores obtained in the first evaluation. It is important to note that, even the level of social withdrawal behavior in this sample was low (*M* = 2.38, *SD* = 2.97), 15.9% of the infants was socially withdrawn, of which 3.5% showed severe social withdrawal behavior (total scores greater than 10).

Table 3.22. *Frequencies of the total scores of the ADBB scale in the first evaluation (T0)*

ADBB T0 Total score	Frequency	Valid percentage
0	48	33.1
1	24	16.6
2	22	15.2
3	18	12.4
4	10	6.9
5	**7**	**4.8**
6	**2**	**1.4**
7	**4**	**2.8**
8	**1**	**0.7**
9	**2**	**1.4**
10	**2**	**1.4**
11	**3**	**2.1**
12	**1**	**0.7**
17	**1**	**0.7**
Total	145	100.0
Missing	0	
Total	145	

Similarly, in the second evaluation, 10.6% of the infants was socially withdrawn, of which 0.8% showed a score corresponding to severe withdrawal behavior (>10) (see table 3.23).

Table 3.23. *Frequencies of the total scores of the ADBB scale in the second evaluation (T1)*

ADBB T1 Total score	Frequency	Valid percentage
0	52	42.3
1	26	21.1
2	16	13.0
3	10	8.1
4	6	4.9
5	**8**	**6.5**
6	**2**	**1.6**
7	**2**	**1.6**
12	**1**	**0.8**
Total	123	100.0
Missing	22	
Total	145	

3.2.2. Infant social withdrawal behavior (m-ADBB)

The descriptive statistics of the total scores of the m-ADBB scale in both assessments are detailed in table 3.24. The theoretical amplitude of the response scale is from 0 to 10, with a cut-off score of two. Hence, m-ADBB total scores above two would indicate the presence of infant social withdrawal behavior. As can be noted, the means of the total scores were below the cut-off in both evaluations. That is to say, the level of social withdrawal behavior was reduced in this sample of infants with CL/P, at T0 and T1. Moreover, the mean of the total scores was higher in the first evaluation (*M* = 1.67, *SD* = 1.97) compared to the second one (*M* = 1.01, *SD* = 1.34). Concerning the distribution of the data, the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.24. Main descriptive statistics of the m-ADBB scale in the first (T0) and the second (T1) evaluation

		m-ADBB T0 Total score	m-ADBB T1 Total score
N	Valid	136	123
	Missing	9	22
Mean		**1.67**	**1.01**
Median		1.00	1.00
Mode		0	0
Standard deviation		1.97	1.34
Asymmetry		1.77	1.61
Std. Error Asymmetry		0.208	0.218
Kurtosis		4.16	2.92
Std. Error Kurtosis		0.413	0.433
Minimum		0	0
Maximum		11	7
Percentile	Percentile 25	0	0
	Percentile 50	1	1
	Percentile 75	2.75	2.00

Regarding the frequencies, in the first evaluation, 24.9% of the infants having CL/P was socially withdrawn (see table 3.25), according to the m-ADBB scale.

Table 3.25. *Frequencies of the total scores of the m-ADBB scale in the first evaluation (T0)*

m-ADBB T0 Total score	Frequency	Valid percentage
0	50	36.8
1	25	18.4
2	27	19.9
3	**18**	**13.2**
4	**4**	**2.9**
5	**5**	**3.7**
6	**1**	**0.7**
7	**4**	**2.9**
8	**1**	**0.7**
11	**1**	**0.7**
Total	136	100.0
Missing	9	
Total	145	

Table 3.26 detailed the frequencies of total scores obtained in the m-ADBB in the second evaluation. As can be noted, 13.8% of the infants showed signs of social withdrawal behavior. Thus, the percentage of social withdrawal behavior decreased from the first to the second evaluation.

Table 3.26. *Frequencies of the total scores of the m-ADBB scale in the second evaluation (T1)*

m-ADBB T1 Total score	Frequency	Valid percentage
0	61	49.6
1	29	23.6
2	16	13.0
3	**9**	**7.3**
4	**6**	**4.9**
5	**1**	**0.8**
7	**1**	**0.8**
Total	123	100.0
Missing	22	
Total	145	

3.2.3. Quality of the parent-infant relationship (PIPE)

Table 3.27 shows the main descriptive statistics of the total scores for PIPE at four (T0) and twelve months (T1). The theoretical amplitude of the response scale is from 0 to 15. The lower scores reflect the more adaptive interactions. As can be seen, the mean of the total score values for the first evaluation were higher (*M* = 2.19, *SD* = 2.90) than for the second one (*M* = 1.51, *SD* = 2.12). That is, the quality of the parent-infant interaction was lower at four months. In addition, the amplitude of the total scores was equal in the first and in the second evaluation. Moreover, the results revealed that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.27. *Main descriptive statistics of the PIPE scale in the first (T0) and the second (T1) evaluation*

		PIPE T0 Total score	PIPE T1 Total score
N	Valid	123	118
	Missing	22	27
Mean		**2.19**	**1.51**
Median		1.00	1.00
Mode		0	0
Standard deviation		2.90	2.12
Asymmetry		1.69	1.99
Std. Error Asymmetry		0.218	0.223
Kurtosis		2.109	4.783
Std. Error Kurtosis		0.433	0.442
Minimum		0	0
Maximum		11	11
Percentile	Percentile 25	0	0
	Percentile 50	1	1
	Percentile 75	3	2

Regarding the frequencies of the PIPE total scores for the first evaluation (T0), it was observed that 85.4% of the interactions were scored as highly adaptive (<5), 9.8% were assessed as slightly adaptive (5 - 9), and only 4.8% were assessed as problematic (≥10). In the second evaluation, 93.2% of interactions were assessed as highly adaptive (<5), 5.2% as slightly adaptive (5-9), while only 1.6% were classified as problematic (≥10).

3.2.4. Impact on family (IOFS)

The results of the descriptive analysis of the total scores of the IOFS are detailed below. It should be mentioned that the theoretical range of the response scale is from 15 to 60, the higher scores reflect a greater impact of the malformation in the family. Table 3.28 shows that, in average, the level of impact on family reported by both parents was low-moderate. In addition, at four months, the impact was greater than at twelve months, in both mothers and fathers. Moreover, mothers' averages were higher than those of fathers.

In relation to the amplitude of the total scores, it was higher in the first evaluation (T0), compared to the second measurement (T1), in mothers and fathers. Furthermore, the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.28. *Main descriptive statistics of the IOFS scale in the first (T0) and the second (T1) evaluation*

		IOFS Mothers T0	IOFS Mothers T1	IOFS Fathers T0	IOFS Fathers T1
N	Valid	137	119	121	100
	Missing	8	26	24	45
Mean		**23.99**	**21.36**	**23.56**	**20.59**
Median		22.00	19.00	23.00	19.00
Mode		15	15[a]	15	15
Standard deviation		6.82	5.81	6.50	5.305
Asymmetry		0.793	1.147	0.785	0.784
Std. Error Asymmetry		0.207	0.222	0.220	0.241
Kurtosis		0.298	0.825	1.388	-0.591
Std. Error Kurtosis		0.411	0.440	0.437	0.478
Minimum		15	15	15	15
Maximum		48	43	51	32
Percentile	Percentile 25	19.00	17.00	17.00	16.00
	Percentile 50	22.00	19.00	23.00	19.00
	Percentile 75	29.00	25.00	29.00	24.00

[a] There are more than one mode value. The smallest value is displayed.

As can be observed in table 3.28, 75% of the mothers presented scores ≤ 29 (interquartile range 19-29) in the first measurement, and scores ≤ 25 (interquartile range 17-25) in the second evaluation. It would indicate a higher impact on mothers at four months, compared to twelve months. It should be noted that at T0 there were eight missing values, corresponding to 5.5% of the sample, while at T1, the missing values were 26 (17.9%).

Concerning the fathers, in the first evaluation, 75% scored ≤ 29 (interquartile range 17-29), and this percentage of fathers scored ≤ 24 (interquartile range 16-24) in the second evaluation. Similar to mothers, the impact on fathers decreased at twelve months. It should be noted that at four months there were 16.6% of missing values and at 12 months, 31%. These findings revealed the importance of psychological support to both parents during the first months after the birth of an infant with CL/P.

3.2.5. Psychological distress (IDP)

Table 3.29 details the main descriptive statistics of the total scores of the Psychological Distress Index (IDP) in both periods of evaluation for mothers and fathers. It should be mentioned that the theoretical range of the response scale is from 14 to 28, the higher scores reflect a greater psychological distress on parents.

As can be noted, the mean of the IDP total scores revealed a moderate level of psychological distress in both parents. Moreover, the mean of psychological distress in mothers was higher than in fathers, in both evaluations. That is, the psychological distress in the mothers is greater than in fathers, at four and at twelve months. Furthermore, the amplitude of the total scores was higher at T1. Moreover, the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.29. *Main descriptive statistics of the IDP scale in the first (T0) and the second (T1) evaluation*

		IDP Mothers T0	IDP Mothers T1	IDP Fathers T0	IDP Fathers T1
N	Valid	135	119	123	102
	Missing	10	26	22	43
Mean		**19.08**	**18.93**	**17.85**	**17.19**
Median		19.00	19.00	17.00	16.50
Mode		16	16	14	14
Standard deviation		3.207	3.369	3.324	3.020
Asymmetry		0.204	0.443	0.683	0.618
Std. Error Asymmetry		0.209	0.222	0.218	0.239
Kurtosis		-0.941	-0.428	-0.438	-0.788
Std. Error Kurtosis		0.414	0.440	-0.433	0.474
Minimum		14	14	14	14
Maximum		26	28	26	25
Percentile	Percentile 25	16.00	16.00	15.00	14.00
	Percentile 50	19.00	19.00	17.00	16.50
	Percentile 75	22.00	21.00	20.00	20.00

Regarding the frequencies of the total scores of the IDP, it was observed that 75% of the mothers scored ≤ 22 (interquartile range 16-22) at T0, and this same percentage of mothers obtained scores ≤ 21 (interquartile range 16-21) at T1. That is, maternal psychological distress was very similar in both measurements. It should be mentioned that in the first evaluation there were 10 missing values (6.9%), while in the second, 17.9% of the sample was missing.

Concerning the fathers, 75% scored ≤ 20 (interquartile range 15-20, interquartile range 14-20) in both evaluations. It should be noted that in the first evaluation, there were 22 missing values (15.2%), while in the second measurement 29.7% of the sample was missing. As can be seen, the total scores of psychological distress in the fathers were lower than those in the mothers, in both evaluations.

3.2.6. Postnatal depression (EPDS)

Table 3.30 shows the results of the descriptive analysis of the total scores of the Edinburgh Postnatal Depression Scale (EPDS), in both evaluations, for mothers and fathers. The theoretical range of the response scale is from 0 to 30, the highest scores indicate the presence of major depression symptoms, with a cut-off score of ten.

As can be observed, the means of the total score showed a low level of depressive symptoms reported by both parents. Additionally, mothers reported higher means than fathers, in both evaluations. It would indicate that mothers having an infant with CL/P usually had greater symptoms of postpartum depression, compared to fathers. It should be noted that the amplitude of the total scores was higher in the mothers, at T0. Moreover, the analysis revealed that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.30. *Main descriptive statistics of the EPDS scale in the first (T0) and the second (T1) evaluation*

		EPDS Mothers T0	EPDS Mothers T1	EPDS Fathers T0	EPDS Fathers T1
N	Valid	131	118	117	92
	Missing	14	27	28	53
Mean		**7.11**	**6.86**	**5.41**	**4.28**
Median		6.00	6.00	5.00	3.00
Mode		5	2[a]	0	0
Standard deviation		4.635	4.84	4.361	4.256
Asymmetry		1.147	0.867	1.103	1.270
Std. Error Asymmetry		0.212	0.223	0.224	0.251
Kurtosis		2.00	0.989	1.630	1.305
Std. Error Kurtosis		0.420	0.442	0.444	0.498
Minimum		0	0	0	0
Maximum		26	24	23	18
Percentile	Percentile 25	4.00	3.00	2.00	1.00
	Percentile 50	6.00	6.00	5.00	3.00
	Percentile 75	9.00	10.00	7.00	6.00

[a] There are more than one mode value. The smallest value is displayed.

In terms of percentages, in the first evaluation, 23.7% of the mothers obtained total scores that indicated the presence of depressive symptoms (EPDS ≥ 10), whereas in the second evaluation, this percentage increased to 28%. In other words, at T1, the percentage of mothers with depressive symptoms was greater than at T0. It is worth mentioning that, in the first evaluation, there were 14 missing values, equivalent to 9.7% of the sample, while in the second evaluation, there were 27 (18.6%).

As regards the fathers, at four months, 16.2% had a total score above the cut-off (EPDS ≥ 10), this percentage decreased to 12% at twelve months. That is, in the second evaluation, the percentage of fathers with symptoms of depression decreased. In addition, in the first measurement, there were 28 missing values (19.3%) for fathers, while in the second evaluation 53 values were missing, equivalent to 36.6% of the sample.

This is to say, at T1, the percentage of depressed mothers increased, meanwhile the percentage of depressed fathers decreased. In addition, in both evaluations, a higher percentage of mothers reported depressive symptoms, in comparison to fathers.

3.2.7. Marital satisfaction (DAS)

Table 3.31 details the results of the descriptive analysis of the total scores of the Dyadic Adjustment Scale (DAS), for both mothers and fathers, at T0 and T1. It should be mentioned that the theoretical range of the response scale is from 16 to 96, the highest scores indicate greater marital satisfaction, with a cut-off of 54.

As can be seen, the total score averages are above this cut-off. That is, both mothers and fathers having an infant with CL/P reported high marital satisfaction, at four and at twelve months. In addition, the results indicated that the absolute values of asymmetry and kurtosis were within the ranges of normal distribution.

Table 3.31. *Main descriptive statistics of the DAS scale in the first (T0) and the second (T1) evaluation*

		DAS Mothers T0	DAS Mothers T1	DAS Fathers T0	DAS Fathers T1
N	Valid	129	105	122	98
	Missing	16	40	23	47
Mean		**73.94**	**72.90**	**74.43**	**73.48**
Median		75.00	75.00	75.00	73.50
Mode		81[a]	70	81	72[a]
Standard deviation		10.199	11.513	10.000	10.908
Asymmetry		-0.787	-0.932	-0.437	-0.580
Std. Error Asymmetry		0.213	0.236	0.219	0.244
Kurtosis		1.493	1.286	0.187	1.008
Std. Error Kurtosis		0.423	0.467	0.435	0.483
Minimum		32	30	48	39
Maximum		96	95	96	96
Percentile	Percentile 25	68.50	67.50	67.75	66.75
	Percentile 50	75.00	75.00	75.00	73.50
	Percentile 75	81.00	82.00	81.00	81.25

[a] There are more than one mode value. The smallest value is displayed.

Regarding the percentages, in the first evaluation, 3.1% of the mothers scored below the cut-off score (DAS ≤ 54), which corresponds to very low marital satisfaction. In the second evaluation, the percentage of mothers suggesting low marital satisfaction increased to 6.7%. That is, the percentage of mothers with high marital satisfaction decreased at twelve months. The missing values in the first evaluation correspond to 11% of the sample, and to 27.6% in the second period.

In relation to the fathers, it was observed that, in the first evaluation, 4.1% reported low marital satisfaction. This percentage was maintained in the second evaluation. It should be mentioned that at four months, there were 23 missing values equivalent to 15.9% of the sample, while at 12 months, there was 47 missing values (32.4%).

These findings revealed that, in general, both mothers and fathers reported high marital satisfaction during the first year after the birth of an infant with CL/P. However, the percentage of mothers with low marital satisfaction increased from 3.1% to 6.7%, meanwhile the percentage of fathers with low marital satisfaction was stable during the first and the second evaluation.

3.3. Hypothesis verification. Analysis of the infant social withdrawal behavior, the quality of parent-infant relationship, and the psychological impact on parents at T0 and at T1

In order to know whether differences exist in infant social withdrawal behavior, quality of the parent-infant relationship, and psychological impact on parents (impact on family, psychological distress, postpartum depression and marital satisfaction), it was carried out an analysis of variance (ANOVA) for repeated measures. The results of this analysis are presented below.

As can be seen in table 3.32, statistically significant differences were found in infant social withdrawal behavior –assessed through ADBB scale (p = 0.004) and m-ADBB scale (p = 0.002)- between T0 and T1. Infant social withdrawal behavior was significantly higher at four months than at twelve months. In addition, the value of square eta (η^2) indicated that the size of the effect was medium.

Statistically significant differences were also found in the impact of the malformation on the family –reported by the mothers (p < 0.001)- between the first and the second evaluation, with a greater impact at four months. In this case, the value of square eta (η^2) indicated that the size of the effect was large.

Similarly, statistically significant differences were found in the impact of the malformation on the family –reported by the fathers (p < 0.001)- between T0 and T1. In this case, the impact of the malformation on the family reported by the fathers was also greater in the first evaluation, than in the second one. It should be noted that the effect size was large (η^2 = .234).

Likewise, the difference was statistically significant in the paternal psychological distress (p = 0.020), between the first and the second evaluation. Paternal psychological distress was higher in the first evaluation. The value of square eta ($\eta 2$) indicated that the size of the

effect was medium. Finally, it was observed a statistically significant difference in postpartum depression in fathers (p = 0.003), between T0 and T1. Paternal postpartum depression was greater at four months, than at twelve months. In this case, the size of the effect was medium-large (see table 3.32).

Table 3.32. *Main descriptive statistics at four (T0) and at twelve (T1) months and ANOVA for repeated measures.*

Variables	T0			T1			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	**2.35**	2.95	123	**1.54**	2.02	123	*F* (1, 122) =8.86, ***p* = .004**, **η^2 = .068**
Infant social withdrawal (m-ADBB)	**1.62**	1.99	119	**1.03**	1.36	119	*F* (1, 118) =10.03, ***p* = .002**, **η^2 = .078**
Parent-infant interaction	2.17	2.90	109	1.63	2.16	109	*F* (1, 108) = 3.37, *p* = .069, η^2 = .030
Impact on Family (mother)	**23.93**	7.06	113	**21.50**	5.90	113	*F* (1, 112) = 25.88, ***p* < .001**, **η^2 = .188**
Impact on Family (father)	**23.75**	6.67	92	**20.49**	5.21	92	*F* (1, 91) = 27.80, ***p* < .001**, **η^2 = .234**
Psychological distress (mother)	19.17	3.16	112	18.97	3.37	112	*F* (1, 111) = 0.50, *p* = .480, η^2 = .005
Psychological distress (father)	**17.91**	3.25	96	**17.27**	3.03	96	*F* (1, 95) = 5.63, ***p* = .020**, **η^2 = .056**
Postpartum depression (mother)	7.06	4.68	108	6.75	4.80	108	*F* (1, 107) = 0.70, *p* = .403, η^2 = .007
Postpartum depression (father)	**5.48**	3.85	80	**4.20**	4.09	80	*F* (1, 79) = 9.68, ***p* = .003**, **η^2 = .109**
Marital satisfaction (mother)	73.52	9.73	100	72.98	11.52	100	*F* (1, 99) = 0.47 *p* = .496, η^2 = .005
Marital satisfaction (father)	73.84	9.33	91	73.45	10.91	91	*F* (1, 90) = 0.21, *p* = .645, η^2 = .002

3.4. Hypothesis verification. Analysis of the type of malformation: CL or CLP

In this section it is detailed the results of the analysis of variance (ANOVA) performed between the groups according to the type of malformation (CL: cleft lip, CLP: cleft lip and cleft palate) with respect to the measured variables of infant social withdrawal behavior, quality of the parent-infant relationship, impact of the malformation on the family, psychological distress on parents, postpartum depression and marital satisfaction.

Table 3.33 shows the main descriptive statistics and the results of the analysis of variance between the group of infants with cleft lip (CL) and the group of infants with cleft lip and palate (CLP), at four months (T0). Statistically significant differences were found in the assessment of impact on family reported by the mothers (p = 0.009), impact on family reported by the fathers (p = 0.008), and marital satisfaction reported by the mothers (p = 0.035). In all cases, the value of square eta (η^2) indicated that the size of the effect was medium.

As can be observed, at four months, the impact of the malformation on the family was higher in the CLP group than in the CL group, in both mothers and fathers. Besides, marital satisfaction reported by the mothers was higher in the CL group, compared to the CLP group. No differences were found in infant's social withdrawal behavior, neither in quality of the parent-infant relationship, between the CLP and the CL groups.

Table 3.33. *Main descriptive statistics and ANOVA between the groups of the type of malformation (CL/CLP) at four months (T0)*

Variables	T0						*F* (ANOVA)
	Cleft Lip			Cleft Lip and Palate			
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	2.39	3.18	23	2.56	3.05	94	*F* (1, 115) = 0.06, *p* = .810, η^2 = .001
Infant social withdrawal (m-ADBB)	1.67	2.13	21	1.82	2.02	89	*F* (1, 108) = 0.010, *p* = .757, η^2= .001
Parent-infant interaction	1.70	2.49	20	2.31	2.96	80	*F* (1, 98) = 0.72, *p* = .397, η^2 = .007
Impact on Family (mother)	**20.64**	4.03	22	**24.62**	6.76	89	*F* (1, 109) = 6.98, ***p* = .009, η^2 = .060**
Impact on Family (father)	**19.79**	4.66	19	**24.14**	6.62	79	*F* (1, 96) =7.29, ***p* = .008, η^2 = .071**
Psychological distress (mother)	18.22	3.28	23	19.41	3.19	87	*F* (1, 108) =2.52, *p* = .116, η^2 = .023
Psychological distress (father)	16.85	2.45	20	18.24	3.40	79	*F* (1, 97) =2.94, *p* = .089, η^2 = .029
Postpartum depression (mother)	6.81	5.69	21	7.28	4.64	85	*F* (1, 104) =0.16, *p* = .691, η^2 = .002
Postpartum depression (father)	5.95	5.60	19	5.28	4.16	74	*F* (1, 91) =0.33, *p* = .566, η^2 = .004
Marital satisfaction (mother)	**77.33**	6.94	21	**72.01**	10.85	84	*F* (1, 103) =4.65, ***p* = .035, η^2 = .042**
Marital satisfaction (father)	76.28	8.68	18	73.37	10.23	79	*F* (1, 95) =1.25, *p* = .267, η^2 = .013

Table 3.34 shows the main descriptive statistics and the results of the analysis of variance between the group of infants with CL and the group of infants with CLP, at twelve months (T1). It was observed a statistically significant difference in the impact on family –reported by the mothers- between the CL group and the CLP group. The impact on the family reported by the mothers in the CLP group was higher than in the CL group. In addition, the value of square eta (η^2) indicated that the size of the effect was medium.

Table 3.34. *Main descriptive statistics and ANOVA between the groups of the type of malformation (CL/CLP) at twelve months (T1)*

Variables	T1						
	Cleft Lip			Cleft Lip and Palate			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	1.21	1.93	19	1.70	2.08	79	F (1, 96) = 0.86, p = .358, η^2 = .009
Infant social withdrawal (m-ADBB)	0.79	1.08	19	1.09	1.42	78	F (1, 95) = 0.75, p = .390, η^2 = .008
Parent-infant interaction	1.56	2.14	18	1.51	2.30	74	F (1, 90) = 0.01, p = .944, η^2 < .001
Impact on Family (mother)	**18.33**	3.23	18	**22.07**	6.32	75	F (1, 91) = 5.87, ***p* = .017**, **η^2 = .061**
Impact on Family (father)	18.50	4.43	14	21.23	5.31	62	F (1, 74) = 3.17, p = .079, η^2 = .041
Psychological distress (mother)	19.00	3.19	18	18.89	3.48	76	F (1, 92) = 0.01, p = .907, η^2 < .001
Psychological distress (father)	16.86	2.62	14	17.67	3.19	64	F (1, 76) = 0.79, p = .377, η^2 = .010
Postpartum depression (mother)	5.29	3.56	17	7.13	5.25	77	F (1, 92) = 1.88, p = .174, η^2 = .020
Postpartum depression (father)	3.50	3.92	12	4.80	4.43	60	F (1, 70) = 0.89, p = .349, η^2 = .013
Marital satisfaction (mother)	76.15	7.01	13	72.46	12.45	70	F (1, 81) = 1.07, p = .303, η^2 = .013
Marital satisfaction (father)	75.69	9.50	13	72.78	10.89	64	F (1, 75) = 0.80, p = .373, η^2 = .011

3.5. Hypothesis verification. Analysis of the laterality of the malformation: unilateral or bilateral

The results of the analysis of variance (ANOVA) carried out between the groups according to the laterality of the malformation (unilateral or bilateral) are detailed below, in relation to the variables of infant social withdrawal behavior, quality of the parent-infant relationship, impact of the malformation on the family, psychological distress on parents, postpartum depression on parents and marital satisfaction.

Table 3.35 shows the main descriptive statistics and the results of the analysis of variance between the groups of infants with unilateral and bilateral malformation, at four months (T0). It was found a statistically significant difference in marital satisfaction reported by the fathers (p = 0.013). Thus, marital satisfaction was higher in the group of fathers having an infant with bilateral malformation. The value of square eta (η^2) indicated that the size of the effect was medium.

Table 3.35. *Main descriptive statistics and ANOVA between the groups of the laterality of the malformation (unilateral or bilateral) at four months (T0)*

Variables	T0						
	Unilateral			Bilateral			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	2.39	2.80	93	3.08	3.94	24	F (1, 115) = 0.98, p = .324, η^2 = .008
Infant social withdrawal (m-ADBB)	1.72	1.91	86	2.04	2.44	24	F (1, 108) = 0.47, p = .497, η^2 = .004
Parent-infant interaction	2.27	2.87	78	1.91	2.94	22	F (1, 98) = 0.27, p = .607, η^2 = .003
Impact on Family (mother)	23.38	6.61	89	25.64	5.81	22	F (1, 109) = 2.14, p = .146, η^2 = .019
Impact on Family (father)	23.72	6.78	76	21.82	5.29	22	F (1, 96) = 1.47, p = .228, η^2 = .015
Psychological distress (mother)	19.25	3.31	88	18.82	2.97	22	F (1, 108) = 0.31, p = .578, η^2 = .003
Psychological distress (father)	17.99	3.18	77	17.86	3.61	22	F (1, 97) = 0.02, p = .877, η^2 < .001
Postpartum depression (mother)	7.30	5.03	83	6.78	4.19	23	F (1, 104) = 0.20, p = .652, η^2 = .002
Postpartum depression (father)	5.49	4.42	74	5.16	4.75	19	F (1, 91) = 0.08, p = .777, η^2 = .001
Marital satisfaction (mother)	73.88	10.28	85	69.65	10.37	20	F (1, 103) = 2.74, p = .101, η^2 = .026
Marital satisfaction (father)	**72.56**	10.03	75	**78.50**	8.50	22	F (1, 95) = 6.36, **p = .013, η^2 = .063**

Concerning the second evaluation, table 3.36 shows the main descriptive statistics and the results of the analysis of variance between the groups of infants with unilateral and bilateral malformation, at 12 months (T1). The results indicated that there were no statistically significant differences between groups. Besides, the size of the effect (η^2) in all cases was small ($\eta^2 \leq .005$), except for marital satisfaction in fathers, in which the size of the effect was between small-medium ($\eta^2 = 0.037$).

Table 3.36. *Main descriptive statistics and ANOVA between the groups of the laterality of the malformation (unilateral or bilateral) at twelve months (T1)*

Variables	T1						
	Unilateral			Bilateral			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	1.67	2.16	78	1.35	1.56	20	F (1, 96) = 0.38, p = .541, η^2= .004
Infant social withdrawal (m-ADBB)	1.06	1.38	77	0.90	1.29	20	F (1, 95) = 0.23, p = .631 η^2 = .002
Parent-infant interaction	1.59	2.32	74	1.22	2.01	18	F (1, 90) = 0.39, p = .534, η^2 = .004
Impact on Family (mother)	21.51	6.12	74	20.68	5.72	19	F (1, 91) = 0.28, p = .595, η^2 = .003
Impact on Family (father)	20.90	5.53	59	20.12	4.19	17	F (1, 74) = 0.29, p = .592, η^2 = .004
Psychological distress (mother)	19.04	3.60	74	18.45	2.64	20	F (1, 92) = 0.47, p = .496, η^2 = .005
Psychological distress (father)	17.47	3.20	60	17.72	2.82	18	F (1, 76) = 0.09, p = .761, η^2 = .001
Postpartum depression (mother)	6.87	5.33	75	6.53	3.64	19	F (1, 92) = 0.07, p = .793, η^2 = .001
Postpartum depression (father)	4.67	4.74	58	4.21	2.15	14	F (1, 70) = 0.12, p = .727, η^2 = .002
Marital satisfaction (mother)	73.12	11.86	65	72.72	11.97	18	F (1, 81) = 0.02, p = .900, η^2 < .001
Marital satisfaction (father)	72.18	10.86	60	77.12	9.22	17	F (1, 75) = 2.90, p = .093, η^2 = .037

3.6. Hypothesis verification. Analysis of the timing of the diagnosis: prenatal or postnatal

The results of the analysis of variance (ANOVA) carried out between the groups according to the timing of the diagnosis (prenatal or postnatal) are detailed below, in relation to the variables of infant social withdrawal behavior, quality of the parent-infant relationship, impact of the malformation on the family, psychological distress of parents, postpartum depression of the parents and marital satisfaction.

Table 3.37 shows the main statistics and the results of the analysis of variance between the groups of infants with prenatal and postnatal diagnosis, in the first evaluation (T0). Results revealed a statistically significant difference in the impact of the malformation on the family – reported by the mothers (p = 0.023) and the fathers (p = 0.038)- between the prenatal and postnatal groups. As can be observed, in this first evaluation, the impact on family was higher in the group of prenatal diagnosis, for both mothers and fathers. In both cases, the value of square eta (η^2) indicated that the size of the effect was small-medium.

Table 3.37. *Main descriptive statistics and ANOVA between the groups of prenatal and postnatal diagnosis at four months (T0)*

Variables	T0						*F* (ANOVA)
	Postnatal			Prenatal			
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	3.05	3.53	19	2.50	3.00	101	F (1, 118) = 0.52, p = .473, η^2 = .004
Infant social withdrawal (m-ADBB)	1.94	2.34	18	1.82	1.98	94	F (1, 110) = 0.06, p = .812, η^2 = .001
Parent-infant interaction	2.21	2.99	19	2.42	3.09	84	F (1, 101) = 0.07, p = .793, η^2 = .001
Impact on Family (mother)	**20.44**	4.82	18	**24.25**	6.68	95	F (1, 111) = 5.29, ***p* = .023, η^2 = .046**
Impact on Family (father)	**19.79**	5.28	14	**23.67**	6.581	86	F (1, 98) = 4.41, ***p* = .038, η^2 = .043**
Psychological distress (mother)	18.72	3.19	18	19.18	3.18	93	F (1, 109) = 0.32, p = .576, η^2 = .003
Psychological distress (father)	19.07	3.22	14	17.78	3.22	87	F (1, 99) = 1.92, p = .168, η^2 = .019
Postpartum depression (mother)	6.81	6.16	16	6.98	4.64	92	F (1, 106) = 0.02, p = .901, η^2 < .001
Postpartum depression (father)	7.07	3.91	14	5.15	4.49	80	F (1, 92) = 2.25, p = .137, η^2 = .024
Marital satisfaction (mother)	76.60	9.56	15	72.86	10.29	90	F (1, 103) = 1.73, p = .191, η^2 = .017
Marital satisfaction (father)	77.85	6.38	13	74.08	10.19	86	F (1, 97) = 1.67, p = .200, η^2 = .017

Moreover, table 3.38 details the main statistics and the results of the analysis of variance between the groups of infants with prenatal and postnatal diagnosis, in the second evaluation (T1). At 12 months, a statistically significant difference in the quality of the parent-infant interaction was found (p = 0.043). A higher quality of the parent-infant relationship was observed in the prenatal diagnosis group, in comparison to the postnatal diagnosis group. The value of square eta (η^2) indicated that the size of the effect was small-medium.

Table 3.38. *Main descriptive statistics and ANOVA between the groups of prenatal and postnatal diagnosis at twelve months (T1)*

Variables	T1						
	Postnatal			Prenatal			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	1.39	2.17	18	1.68	2.04	82	F (1, 98) = 0.30, p = .587, η^2 = .003
Infant social withdrawal (m-ADBB)	0.72	1.07	18	1.11	1.41	82	F (1, 98) = 1.21, p = .274, η^2 = .012
Parent-infant interaction	**2.47**	3.08	19	**1.32**	1.93	76	F (1, 93) = 4.20, ***p* = .043, η^2 = .043**
Impact on Family (mother)	19.53	3.96	19	21.68	6.30	77	F (1, 94) = 2.00, p = .161, η^2 = .021
Impact on Family (father)	19.85	5.58	13	20.76	5.13	66	F (1, 77) = 0.33, p = .566, η^2 = .004
Psychological distress (mother)	18.47	2.71	19	19.00	3.57	78	F (1, 95) = 0.36, p = .550, η^2 = .004
Psychological distress (father)	16.31	2.13	13	17.74	3.17	68	F (1, 79) = 2.41, p = .124, η^2 = .030
Postpartum depression (mother)	6.21	4.00	19	6.81	5.26	77	F (1, 94) = 0.21, p = .647, η^2 = .002
Postpartum depression (father)	4.58	4.66	12	4.70	4.40	63	F (1, 73) = 0.01, p = .935, η^2 < .001
Marital satisfaction (mother)	76.43	9.77	14	72.41	11.98	71	F (1, 83) = 1.39, p = .242, η^2 = .016
Marital satisfaction (father)	74.33	10.82	12	73.19	10.77	67	F (1, 77) = 0.11, p = .737, η^2 = .001

3.7. Hypothesis verification. Analysis of the waiting time prior to the first repair surgery: early or late surgery

This section details the results of the analysis of variance (ANOVA) performed between the groups according to the waiting time before the first repair surgery (early: 0-90 days, late: more than 90 days), in relation to the variables of infant social withdrawal behavior, quality of the parent-infant relationship, impact of the malformation on the family, psychological distress on parents, postpartum depression, and marital satisfaction.

Table 3.39 shows the main descriptive statistics and the results of the analysis of variance between the groups of early and late surgery in the first evaluation (T0). It was observed statistically significant differences in the impact of the malformation on the family –reported by the mothers ($p < 0.001$) and the fathers ($p < 0.001$)- between the early and the late surgery groups. As shown in table 3.39, the impact of the malformation on the family was greater in the late surgery group, in comparison with the early surgery group, for both mothers and fathers. In both cases, the square eta value (η^2) indicated that the effect size was large.

Furthermore, it was found a statistically significant difference in postpartum depression in the mothers ($p = 0.024$). That is, maternal depressive symptoms were greater in the late surgery group, as compared to the early surgery group. In this case, the value of square eta (η^2) indicated that the size of the effect was medium.

Table 3.39. *Main descriptive statistics and ANOVA between the groups of early and late surgery at four months (T0)*

Variables	T0						
	Early surgery			Late surgery			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	2.81	3.77	48	2.48	2.57	64	F (1, 110) = 0.30, p = .586, η^2 = .003
Infant social withdrawal (m-ADBB)	1.85	2.32	47	1.88	1.83	57	F (1, 102) = 0.00, p = .949, η^2 < .001
Parent-infant interaction	2.55	3.52	44	2.33	2.70	52	F (1, 94) = 0.12, p = .732, η^2= .001
Impact on Family (mother)	**20.98**	5.61	43	**25.50**	6.52	62	F (1, 103) = 13.65, **p < .001, η^2 = .117**
Impact on Family (father)	**20.03**	4.89	35	**25.23**	6.94	57	F (1, 90) = 15.01, **p < .001, η^2 = .143**
Psychological distress (mother)	18.49	3.19	45	19.59	3.20	59	F (1, 102) = 3.04, p = .084, η^2 = .029
Psychological distress (father)	17.46	3.22	37	18.19	3.21	57	F (1, 92) = 1.17, p = .283, η^2 = .013
Postpartum depression (mother)	**5.71**	4.87	42	**7.93**	4.72	58	F (1, 98) = 5.28, **p = .024, η^2 = .051**
Postpartum depression (father)	4.50	3.96	34	5.77	4.21	52	F (1, 84) = 1.95, p = .166, η^2 = .023
Marital satisfaction (mother)	73.26	10.48	39	72.57	10.80	58	F (1, 95) = 0.10, p = .757, η^2 = .001
Marital satisfaction (father)	76.68	7.96	34	72.53	10.74	57	F (1, 89) = 3.81, p = .054, η^2 = .041

At 12 months, there were statistically significant differences in the impact of the malformation on the family –reported by the mothers (p = 0.007) and the fathers (p = 0.009)- between the early and late surgery groups. It should be observed that, the impact of the malformation was greater in the late surgery group (see table 3.40), for both mothers and fathers. In addition, the square eta value (η^2) indicated that the size of the effect was medium in both cases.

Moreover, there was a statistically significant difference in psychological distress reported by the fathers ($p < 0.001$), which was higher in the late surgery group. In this case, the square eta value (η^2) indicated that the effect size was large.

Finally, we also found statistically significant differences in the postpartum depression –reported by the mothers ($p = 0.012$) and the fathers ($p < 0.001$)- between the early and late surgery groups. The postnatal depression was greater in the late surgery group, in both parents. Also, the value of square eta (η^2) indicated that the size of the effect was medium for the mothers, and large for the fathers.

Table 3.40 *Main descriptive statistics and ANOVA between the groups of early and late surgery at twelve months (T1)*

Variables	T1						
	Early surgery			Late surgery			*F* (ANOVA)
	M	*SD*	*n*	*M*	*SD*	*n*	
Infant social withdrawal (ADBB)	1.50	1.77	42	1.65	1.85	51	*F* (1, 91) = 0.50, *p* = .698, η^2 = .002
Infant social withdrawal (m-ADBB)	1.00	1.15	42	1.02	1.32	51	*F* (1, 91) = 0.01, *p* = .940, η^2 < .001
Parent-infant interaction	1.71	2.53	42	1.59	2.08	46	*F* (1, 86) = 0.07, *p* = .797, η^2 = .001
Impact on Family (mother)	**19.50**	4.82	40	**22.90**	6.46	49	*F* (1, 87) = 7.59, ***p* = .007, η^2 = .080**
Impact on Family (father)	**18.75**	4.38	28	**22.04**	5.49	45	*F* (1, 71) = 7.19, ***p* = .009, η^2 = .092**
Psychological distress (mother)	18.20	3.42	41	19.49	3.37	49	*F* (1, 88) = 3.24, *p* = .075, η^2 = .035
Psychological distress (father)	**15.77**	2.12	30	**18.56**	3.04	45	*F* (1, 73) = 18.91, ***p* < .001, η^2 = .206**
Postpartum depression (mother)	**5.33**	3.72	39	**8.04**	5.74	50	*F* (1, 87) = 6.51, ***p* = .012,** η^2 = .070
Postpartum depression (father)	**2.48**	3.06	27	**6.24**	4.70	42	*F* (1, 67) = 13.49, ***p* < .001, η^2 = .168**
Marital satisfaction (mother)	73.82	12.74	33	72.63	10.69	46	*F* (1, 77) = 0.20, *p* = .655, η^2 = .003
Marital satisfaction (father)	76.00	10.51	28	71.93	10.02	45	*F* (1, 71) = 2.74, *p* = .103, η^2 = .037

A brief summary of the findings is presenting below. The **descriptive analysis of the items** of the ADBB and the m-ADBB scales –which assess social withdrawal behavior- showed that vocalizations was the item with greater alterations in both assessments. This finding may be related to the multiple interventions in the oral cavity that infants having CL/P require. Regarding the quality of the parent-infant interaction -evaluated with the PIPE- larger difficulties were found in the item concerning the development of the parent-infant game, and fewer alterations were found at the beginning of the interaction, in both evaluations.

The descriptive analysis of the items of IOFS revealed that –in mothers and fathers- the greatest impact of the CL/P malformation on the family lies in the trips to the hospital, at T0 and T1. This result should be take into account by the medical team in order to reduce, as far as possible, the multiple appointments at the hospital. In this line, home medical care may be beneficial for these families.

In relation to the descriptive analysis of the items of the IDP –which assess psychological distress- both, mothers and fathers reported higher rates of fear at four months, and higher rates of irritability at twelve months. This may be interpreted as an adaptation process of the parents to their infant's malformation. Moreover, our results provide the guidelines to support these parents according to each period.

Regarding the descriptive analysis of the items of the EPDS –which assess postnatal depression- mothers and fathers reported greater feelings of reproach and guilt, at four and at twelve months. Besides, thoughts about self-harm were the less frequent in both assessments. These results highlighted the necessity of psychological support to mothers and fathers having an infant with CL/P, in order to reduce the

feelings of guilt, and to enhance their understanding of the CL/P etiology.

Lastly, concerning the descriptive analysis of the items of the DAS –which assess marital satisfaction- it was observed that divorce was the less common thought between mothers and fathers, at four and at twelve months. In addition, mothers reported lower satisfaction in relation to working on a common project with their partner, while fathers showed less satisfaction regarding the exchange of stimulating ideas between the couple. These results emphasize the aspects that are required to work with the parents as a couple.

Regarding the results of **the analysis of the total scores**, the means of the ADBB total scores revealed that the social withdrawal behavior was low in our sample, in both assessments. However, in terms of percentages, at four months, 15.9% of the infants showed signs of social withdrawal behavior, of which 3.5% corresponded to a severe degree of withdrawal. At twelve months, these percentages decreased to 10.6% and 0.8%, respectively. According to the m-ADBB, 24.9% of the infants was socially withdrawn at four months, and 13.8% at twelve months. These results may indicate that, although the level of social withdrawal behavior was low in this sample of infants with CL/P, the percentages of withdrawn infants correspond to those reported in at risk populations.

As regards the analysis of the PIPE total scores –which measures the quality of the parent-infant relationship- we observed that the mean scores were low, suggesting adaptive interactions in our sample. However, the quality of the parent-infant relationship was lower at T0 than at T1. 4.8% of the parent-infant interactions were rated as problematic in the first measurement, and 1.6% in the second assessment. This indicates the necessity of psychological support to parents and infants during the first months after the birth in order to enhance adaptive patterns of interaction.

In relation to the analysis of the total scores of the IOFS, it was observed a low-moderate impact of the malformation on family life. Besides, the average of the total scores were higher at four months than at twelve months, in both mothers and fathers. This means that the impact of the malformation on the family was greater during the first months after the birth of an infant with CL/P.

In regard to the analysis of the total scores of the IDP, we found a low-moderate level of psychological distress on parents. Additionally, the means of the total scores were higher in mothers, in both evaluations. That is to say, mothers having an infant with CL/P often report greater psychological distress than fathers during the first year postpartum.

Furthermore, descriptive analysis of the EPDS total scores revealed low levels of postnatal depression in parents having an infant with CL/P, during the first year postpartum. In addition, mothers' mean scores were higher in both evaluations. This suggests that mothers having an infant with CL/P had higher depressive symptoms. It is important to say that, although the level of depressive symptoms was low, at four months, 23.7% of the mothers showed signs of postpartum depression, compared to 16.2% of the fathers. At twelve months, the percentage of mothers with signs of depression increased to 28%, and that of the fathers decreased to 12%.

Finally, the analysis of the DAS total scores showed high levels of marital satisfaction in parents having an infant with CL/P. However, it should be noted that the level of marital satisfaction decreased in the second assessment, in both mothers and fathers.

According to the **verification of the general hypothesis**, we observed that the infant social withdrawal behavior, the impact of the malformation on the family –reported by mothers and fathers-, as well as paternal psychological distress and postpartum depression in fathers were significantly higher at T0 than at T1. This is, in general, the

psychological impact of the CL/P malformation is higher in the first months after the birth of an infant with CL/P, in both parents and infants.

Concerning the **verification of the secondary hypothesis**, in the analysis between groups according to the type of malformation (CL or CLP), at four months, the impact of the malformation in the family was significantly higher in mothers and fathers in the CLP group, compared to the CL group. In addition, marital satisfaction was significantly higher in mothers of the CL group, in comparison to the CLP group. At twelve months, it was found a statistically significant difference in the impact of the malformation on the family, reported by the mothers, with a greater impact on the CLP group.

In the analysis between groups according to laterality of the malformation (unilateral or bilateral), at four months, a statistically significant difference was observed in paternal marital satisfaction. That is, marital satisfaction on fathers was higher in the group of bilateral malformation. In the second evaluation, no statistically significant differences were found between laterality malformation groups.

In relation to the analysis between groups according to the type of diagnosis (prenatal or postnatal), at four months, it was observed a statistically significant greater impact of the malformation in the prenatal diagnosis group, in both mothers and fathers. Besides, at twelve months, the quality of the parent-infant interaction was significantly lower in the postnatal diagnosis group, compared to the prenatal diagnosis group.

Regarding the analysis between groups according to the time of the repair surgery (early or late), results revealed statistically significant differences in the impact of the malformation on the family -reported by mothers and fathers-. It was observed a greater impact of the

malformation on the family in the late surgery group. Similarly, maternal depression was significantly higher in the late surgery group. At twelve months, the impact of the malformation on the family and the postpartum depression were significantly higher in the late surgery group, for both mothers and fathers. In addition, psychological distress on fathers was significantly higher in the late surgery group. Thus, an early repair surgery on infants with CL/P enhances the psychological wellbeing of parents.

All these results lead to a better understanding of the psychological state of the infants and their parents in the context of the CL/P malformation, and provide important guidelines for psychological support and intervention. In the next chapter, all these findings will be deeply discussed.

4. DISCUSSION

The current longitudinal and prospective study provides a description of the evolution of the infant social withdrawal behavior, the psychological impact of the malformation on parents, and the quality of the parent-infant relationship, at four (T0) and at twelve months postpartum (T1), in infants with Cleft Lip and Palate (CL/P) and their parents. Moreover, the present research offers interesting information about the differences in these variables between groups according to the type and the laterality of the CL/P malformation, the type of diagnosis and the time of the repair surgery. These findings contribute to a better understanding of the psychological state of the infants and parents in the context of the CL/P malformation, and provide important guidelines for psychological support and intervention.

The discussion of the results will be done in the following order. Firstly, will be analyzed the results related to the general hypothesis –which states differences in the variables between T0 and T1- taking into account the descriptive analyses of each of the variables. Subsequently, we will discuss the outcomes related to the specific hypotheses that indicate differences between the groups. Then, will be presented the conclusions, some practical implications derived from our work, and the strengths and limitations of the current thesis.

According to the **general hypothesis**, it was found that the infant social withdrawal behavior, the impact of the malformation on the family –reported by mothers and fathers- as well as paternal psychological distress, and postpartum depression in fathers, were significantly higher in the first evaluation (T0), compared to the second one (T1). In other words, the psychological impact of the CL/P malformation was greater in the first months postpartum, in both parents and infants.

These findings may be explained through the family stress and coping model (McCubbin, Thompson, & McCubbin, 1996), which states that families having an infant with a medical condition seek to restore balance and progress to a state of long-term adaptation. Thus, in our study, the decrease of the psychological impact on parents and on infants may be understood as a major adaptation to the context of the CL/P malformation.

Particularly, the fact that the infants with CL/P were significantly more **socially withdrawn** at four months postpartum –in comparison to twelve months- may be due to the multiple intrusive medical treatments that are carried out, in the first months, in order to facilitate the feeding and other vital functions. Then, when the repair surgery is performed, the social withdrawal behavior may result as a consequence of the post-surgical pain, since severe pain has been found to be a risk factor for social withdrawal behavior (Gauvain-Piquard, Rodary, Rezvani, & Lemerle, 1987; Gauvain-Piquard, Rezvani, Rodary, & Serbouti, 1999). In addition, at twelve months, infants may develop different and more complex strategies to cope with distress, and also, the support given by the medical team through the development of the current study may had influenced in the decrease of social withdrawal behavior in infants, since the attention given to the infant during the ADBB evaluation represents a first stage of the intervention (e.g., Pérez-Martínez & Guedeney, 2015).

Regarding the analysis of the **total scores of the ADBB** (Alarm Distress Baby Scale), the global level of social withdrawal behavior was low in our sample. However, in terms of percentages, at four months, 15.9% of infants with CL/P were socially withdrawn, while this percentage decreased at 10.6% at twelve months. When the assessment was carried out through the m-ADBB (modified version of the Alarm Distress Baby Scale), the percentage of socially withdrawn infants was 24.9% at four months, and 13.8%, at twelve months. This

is, both scales perceived a decrease in the percentage of social withdrawal behavior from the first to the second assessment.

The discrepancies in the detection of the social withdrawal behavior by each of the scales (ADBB and m-ADBB), may be because the m-ADBB excludes items that are more difficult to evaluate (i.e., self-stimulating gestures), making easier the identification of the alarm signs. Similarly, Guedeney et al. (2013) found that because of its simplified coding and scoring scheme, as compared to the ADBB, the m-ADBB may be more practical for evaluating social withdrawal behavior in vulnerable populations.

Even though the analysis of the total scores revealed that, in general, the level of social withdrawal behavior was low in infants with CL/P, the percentages of socially withdrawn infants correspond to those of the risk population in France. For example, Guedeney, Foucault, Bourgen, Larroque and Mentré (2008) reported a 13%, meanwhile Guedeney et al. (2012) observed a 14%, and Guedeney et al. (2016a) found about 20% infants considered socially withdrawn at age one year, in a risk population. Moreover, the percentage of social withdrawal behavior observed in our sample, at four months, was higher than the 12% of withdrawn infants born very preterm evaluated at 6 months by Cambonie et al. (2017).

Compared to more severe medical conditions, the percentage of social withdrawal behavior is lower in infants with CL/P. For instance, Tauber et al. (2017) observed that 62% of infants –under 6 months- with a genetic diagnosis of Prader-Willi Syndrome was socially withdrawn. It should be noted that our study is the first one in which the social withdrawal behavior is assessed –through the ADBB scale and m-ADBB scale- in infants having Cleft Lip and Palate in the early postpartum period. Our findings emphasize the necessity of the detection of social withdrawal behavior in the first months of life, as a silent signal of

suffering that must be interpreted and treated, especially when infants present a medical condition.

In the line of the **analysis of the items of the ADBB and the m-ADBB scales**, vocalizations was the dimension of social withdrawal with higher difficulties, at four and at twelve months. This is to say, infants with CL/P showed greater lack of vocalizations expressing pleasure (i.e., cooing, laughing, or babbling) but also expressing displeasure or pain (i.e., squealing, screaming, or crying) in both assessments.

These findings are logical if we consider that CL/P malformation affects the oral cavity. The oral structural deficit itself, the post-surgery discomfort, and the use of oral devices –such as palatal obturators- to modify oral structures may complicate the production of vocalizations. The results of our research are consistent with previous studies (Montirosso et al., 2012; Murray et al., 2008) that found that infants with CL/P make fewer communicative signals, and producing fewer positive vocalizations, in comparison with infants with no CL/P.

Furthermore, it has been found that, in terms of functionality, infants with CL/P may experience complications in phonation, hearing and ventilation that also may make difficult the production of vocalizations. In addition, infants having CL/P are more prone to have otitis media, and it has been shown that this medical condition –during the first six months of life- is correlated with delays in pre-linguistic vocal development (Rvachew, Slawinski, Williams, & Green, 1999).

Our findings are in line with previous studies which stated that children with cleft lip and palate have an increased risk for early speech and language delays (Chapman, Hardin-Jones, Schulte, & Halter, 2001; Hardin-Jones & Jones, 2005; Scherer, D'Antonio, & Kalbfleisch, 1999). All these findings emphasize the necessity of assessing social withdrawal behavior, paying especially attention to the vocalizations

item, from the first months of infant's life, in order to orient parents to encourage their infant's oral production.

In this regard, specific sounds are known to be difficult for infants with CL/P to produce because of the functional and structural deviations associated with the cleft malformation. For instance, Chapman et al. (2001) found that babies with cleft palate had smaller canonical babbling ratios than their age-matched peers, with just 57% of the babies with cleft palate reaching the canonical babbling stage by 9 months compared to 93% of the non-cleft babies. Thus, it is coherent that –in our study- the vocalizations is the dimension in which major difficulties have been found.

Concerning the verification of the general hypothesis, no statistically significant difference in the **quality of parent-infant relationship** was observed, between the first and the second evaluation. However, we should take into account that the social withdrawal behavior is considered an alarm signal indicating a lasting difficulty in the parent-infant interaction. In this line, the **analysis of the total scores of the PIPE** (Pediatric Infant Parent Exam) revealed that the total score average was higher in the first assessment, in comparison with the second evaluation. This is to say, at four months –compared twelve months- there were more alterations in the parent-infant interaction, which leads to a lower quality of the relationship.

These results may be explained because, in the case of infants with CL/P, during the first months postpartum, the malformation makes infants less attractive (Goodacre, Hentges, Moss, Short, & Murray, 2004) and for parents result in greater difficulty to interpret their expressions (Field & Vega-Lahr, 1984). In addition, our results showed that the psychological impact of the malformation on parents is greater in the first months after birth. Thus, this psychological distress leads parents to be less responsive and sensitive in the interpretation of their infants' signals (Koomen & Hoeksma, 1992; Montirosso et al., 2012),

and hinders their parental role, which, in turn, increases the probability of alterations in parent-infant relationship. Hence, these descriptive analyses of the total scores of the PIPE suggest a major necessity of psychological support during the early postpartum period, in order to improve the interactional skills in parents and infants in the context of CL/P malformation.

According to the general hypothesis, in our study, it was observed a statistically significant difference in the **psychological impact on parents** –in terms of impact on family, paternal psychological distress, and paternal postnatal depression- between the first and the second evaluation. Our findings confirm that the first months after the birth of an infant with a medical condition were the hardest for mothers and fathers (e.g., Zeytinlogu et al., 2016a). Additionally, adaptation is a particular challenge for parents having an infant with CL/P, since the malformation implies special medical care, and parents should cope with the altered appearance of the baby, which often is associated to negative social feedback (Leemreis et al., 2014). However, this adaptation became easier over time. Thus, the decrease of the psychological impact on parents may be interpreted as a parent's inclination to adapt to the infant's medical condition during the first year.

Impact on family –reported by both parents- was significantly higher in the first evaluation, compared with the second assessment. These findings may indicate that parents perceive more changes in the normative behavior of the family in the first months after the birth of an infant with CL/P. It could be due to the several adaptations that parents should make in their life routines in order to provide –for instance-medical care to their infant. Besides, parents should also cope with feeding difficulties, hospital stays, medical treatments, and additional financial, social, and psychological burden.

The results of the **descriptive analysis of the items of the IOFS** (Impact On Family Scale), the highest impact reported by mothers and fathers –in both assessments- was in terms of travelling to hospital. The medical team should consider this point in order to reduce, as far as possible, the numerous appointments that parents having an infant with CL/P should do. In this line, home medical care may be beneficial for these families.

In addition, the **analysis of the total scores of the IOFS** revealed that the median scores of the IOFS in our sample were higher in comparison with those reported by Boudas et al. (2013) in a sample of parents with a child –aged 6 to 12 years- with CL/P, in France. This indicates that the impact on family is greater during the first year of an infant's with CL/P life, and highlights the importance of evaluating the psychological impact of the malformation on family during the early postnatal period, in order to provide adequate psychological support to parents.

Regarding **psychological distress**, it was observed a statistically significant difference on fathers, between the first and the second assessment. This is to say, psychological distress on fathers was significantly higher at four months, in comparison with at twelve months evaluation. These results may be interpreted as a major adaptation of the fathers to the medical condition of their infants with CL/P.

When we consider the **analysis of total scores of the IDP** (Psychological Distress Index), in terms of percentages, the psychological distress was higher in the first assessment –compared with the second assessment- for both mothers and fathers. The fact that parents were more distressed in the first evaluation may be due to the fact that transition to parenthood is a stressful period as itself, and particularly, because having an infant with a medical condition, in this case with CL/P, add more stressors to the parental roles. Our

findings were consistent with previous studies in the context of congenital anomalies (e.g., Fonseca et al., 2012; Kaasen et al., 2010).

Furthermore, in the analysis of the total scores, the mean score on psychological distress was higher in mothers than in fathers on both evaluations. In other words, the psychological distress on fathers decreased significantly over the first year of an infant's life, while mothers continue in a state of emotional suffering at twelve months postpartum. These findings are in line with the elevated levels of psychological distress in mothers in comparison to fathers found by Doherty et al. (2009) in a study with parents of infants with severe congenital heart disease.

Interestingly, concerning the **analysis of the items of the IDP**, in the first assessment, mothers and fathers scored higher in items related to afraid, while in the second assessment, they scored higher in items referring to irritability. This may be explaining because, during the first months after birth, parents are frightened facing the malformation of their infant, maybe because of the different changes they should cope with, for example in terms of medical care and the expectations related to the repair surgery of the cleft. As well, the greater presence of irritability, at twelve months after birth, may be related to the progress in the grief process for the loss of their imagined baby, in which parents typically experience a variety of emotions including rage, disappointment, anxiety, protectiveness, failure or guilt, and depression (Landsdown, 1981; Slade, Emerson, & Freedlander, 1999). This is an interesting finding that provides guidelines for working with parents in psychotherapy.

In relation to the evaluation of the postnatal depression, only **paternal depressive symptoms** were significantly higher at four months, compared with twelve months evaluation. This finding may indicate a favorable adaptation in fathers to their infant with CL/P. Moreover, the **analysis of the total scores of the EPDS** (Edinburgh Postnatal

Depression Scale) showed that the total scores averages were higher in the first assessment –for both mothers and fathers- in comparison with the second assessment. Similar results were found by Fonseca, Nazaré and Canavarro (2012) who observed that parents of infants with congenital anomalies experienced a significant reduction in depression and anxiety symptoms over time. This is, in our study, most of the parents showed a pattern of recovery from the early postnatal period to twelve months after the birth of an infant with CL/P.

Similar to psychological distress, the fact that the level of depressive symptoms decreases over the time may be also explained because, at birth, the disfigured face of the baby confronts parents with their expectations about their imagined and hoped baby. Then, this confrontation is followed by an adaptation process in which parents typically experience a variety of emotions including rage, disappointment, anxiety, protectiveness, failure or guilt, and depression (Landsdown, 1981; Slade, Emerson, & Freedlander, 1999). This subsequent adaptation is similar to a process of mourning which consists of a first reaction of shock or confusion, followed by denial, sadness and rage, adaptation, and reorganization (Drotar, Baskiewicz, Irvin, Kennell, & Klaus, 1975).

Additionally, in terms of **percentages of postnatal depression**, mothers showed mean scores higher than fathers, in both evaluations. In the first assessment, 23.7% of the mothers were depressed vs. 16.2% of the fathers, while this percentage increased for mothers to 28%, and decreased in fathers to 12%, in the second assessment. In other words, mothers having an infant with CL/P are more likely to present depressive symptoms than fathers, and the percentage of depressed mothers increased over time.

The percentage of depressive mothers, at four months, was comparable to that one reported by Braarud et al. (2013) in mothers with premature infants, at three months postpartum. That is, the

percentage of depressed mothers is similar to perinatal risk populations. However, this percentage is greater than the 10-15% of maternal depression reported in general population during the first year postpartum (e.g., Gavin et al., 2005). Regarding paternal depression, percentages were also higher than the 10.4% reported by Paulson and Bazemore (2010) in the prenatal and the postnatal period. Moreover, our results are in contrast with some studies (e.g., Sank, Berk, Cooper, & Marazita, 2003; Weigl et al., 2005) that affirm that the distribution of depression in parents having children with CL/P is similar to normal population.

Furthermore, our results coincide with previous research reporting that fathers of disabled or with congenital heart diseases infants tend to have lower depression scores than mothers (Bevilacqua et al., 2013; Gray, 2003; Pelchat et al., 1999). Differences in postnatal depression between mothers and fathers may be explained by the greater set of physical and biological changes experienced by mothers during the transition to parenthood, along with their main role as caregivers (Nomaguchi & Milkie, 2003). For example, it has been found that mothers visit their infants more frequently and for longer than fathers, and are more engaged in their infant's care (Franck & Spencer, 2003).

This is, the role of main caregiver, of an infant with a medical condition, may be associated with the increased percentage of maternal depression at twelve months. All these results are in line with a previous study that reported that having an infant with congenital heart disease had prolonged effects on mothers, and there was a significantly larger increase in depression symptoms at 6 and at 18 months after delivery (Solberg et al., 2011). Further studies are needed in order to examine which specific factors are associated with maternal depression one year after the birth of an infant having a CL/P.

Interestingly, the **analysis of the items of the EPDS** showed that mothers and fathers scored higher in the item related to self-blame, in

both assessments. This is to say, the self-blame and the guilt are the most frequent feelings on parents having an infant with CL/P. Similar results were observed in previous studies with mothers (e.g., Baker et al., 2009; Davalbhakta & Hall, 2000) and with fathers (Zeytinoglu et al., 2016a) having an infant with a medical condition. These findings highlight the need to inform parents about the etiology of CL/P, given that parents' beliefs about causation of the malformation have been found to impact their own psychosocial adjustment (Nelson et al., 2009). Thus, parents may benefit from a psychological support in order to reduce these feelings of reproach and guilt.

Concerning **marital satisfaction**, no statistically significant differences were observed between the first and the second evaluation. Nevertheless, the **analysis of the total scores of the DAS** (Dyadic Adjustment Scale) showed that the averages in marital satisfaction were higher in fathers and mothers. In terms of **percentages**, most of the couples reported a high marital satisfaction. At four months, almost 97% of mothers and 96% of fathers reported a good marital satisfaction. At twelve months, this percentage was consistent in fathers, meanwhile the percentage of mothers satisfied with their marital relationship decreased to 93.3%.

These results coincide with previous studies, for instance, Zeytinoglu et al. (2016b) found that most couples reported that raising a child having a CL/P made their relationship stronger because this experience provided an opportunity to work as a team and to be supportive to each other. In this regard, good marital satisfaction can be understood as a sign of adaptation of parents to their infant's medical condition. Moreover, a satisfying relationship and a supportive partner can function as a buffer against distress and hopelessness (Murphy, Christian, Caplin, & Young, 2007). Thus, having an infant with CL/P does not necessarily pose a threat to the parents' marital relationship.

The analysis of the total scores showed that the percentage of mothers with low marital satisfaction increases in the second evaluation. This may be due to the different involvement that mothers and fathers have in the care of their infant with CL/P. Usually, mothers become the main caregiver, while fathers go back to work soon after the birth. Hence, mothers should cope, more often, with the different stressors related to the medical condition of their infant. Additionally, some studies have found that mothers caring for a child with CL/P feel frustrated with the division of labor at home, and they hope their husbands would become more involved (Zeytinoglu et al., 2016b); this may contribute to the decrease of marital satisfaction on mothers, at twelve months.

Moreover, as Favez et al. (2009) stated, the expectations each parent develops toward the other as a mother or father, also play a fundamental role in marital satisfaction. Accordingly, at twelve months, the lack of fulfillment of expectations related to the involvement of fathers in the care of their infant with CL/P may increase the percentage of unsatisfied mothers within their marital relationship. However, this statement should be confirmed in future research. Additionally, at twelve months, the increased percentage of depressed mothers may be associated with the greater percentage of low marital satisfaction on mothers. Further studies are needed to explore this possible association in the context of CL/P malformation.

Regarding the **specific objective** related to exploring the differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the Cleft Lip and Palate (CLP) and Cleft Lip (CL) malformation groups, it was observed that the impact on the family –reported by mothers and fathers- was higher in the CLP group, compared to the CL group, in the first evaluation. Additionally, marital satisfaction on mothers was higher in the CL group, in comparison to the CLP group. In the second

evaluation, only impact on family –as informed by mothers- was significantly higher in the CLP group as compared to the CL group.

These results suggest that having an infant with Cleft Lip and Palate malformation constitutes a major challenge for parents, in comparison to the Cleft Lip malformation. This fact is logical if we take into account that CLP malformation includes more severe implications in infants, than CL malformation. For instance, CLP includes cosmetic problems, but also more serious consequences affecting feeding, hearing, and speech that constitute major difficulties for caring the infant. Furthermore, CLP requires more complex medical treatment, including additional surgeries and several visits to the hospital.

Similar results have been found in previous studies. For example, Slutsky (1969) observed that parents experienced greater shock, hurt, disappointment, and helpless resentment in the group of Cleft Lip and Palate –in comparison with the Cleft Palate group- but both groups were in need of support. In contrast, Nidey et al. (2015) did not find significant differences in parental psychosocial status depending on the type of CL/P malformation. However, this study took into account parents of children aged 0-17 years with oral clefts.

Interestingly, mothers of the CLP group reported significantly lower marital satisfaction, than those of the CL group. This leads to assume that the severity of the infant's malformation is related to the marital satisfaction on mothers. In this line, Dale et al. (2012) observed that the stressors related to the hospital visits, surgeries, financial burden, feeding problems and uncertainties about complications might generate relationship instability in the parental couple. All these stressors are more likely in cases of CLP due to the major medical care requirements. However, our results are in contrast with that found by Despars et al. (2009) in which the complexity of the CL/P did not significantly influence maternal stress. Though, these authors observed

that mothers who reported low posttraumatic stress symptoms, were more disengaged with their infants, or vice versa.

Moreover, at twelve months, the psychological impact on family –reported by the mothers- was also significantly higher in the CLP group, in comparison to the CL group. This means that mothers perceived that changes in the normative behavior of the family –attributable to the infant's medical condition- were still great at twelve months after birth, in the Cleft Lip and Palate group. Hence, these results emphasize that mothers having an infant with CLP –in comparison with CL group- require major necessity of psychological support during the first year of infant's life.

Concerning the **secondary hypothesis** related to exploring the differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the laterality of the malformation, our results revealed that, at four months, fathers having an infant with bilateral malformation reported significantly higher marital satisfaction, in comparison with the unilateral malformation group. These results are consistent with previous studies with parents of infants with congenital heart disease. For example, Dale et al. (2013) suggest that having an infant with a medical condition created in parents a feeling of stronger mutual obligation to stay together and to help each other through a difficult time, which may increase their marital satisfaction. A similar situation could happen to the fathers having an infant with a bilateral cleft, since this malformation is more visible than the unilateral cleft.

In addition, this result may be associated with the fact that fathers often assume the role of provider, while mothers are typically the primary caregiver. This situation may lead to a greater satisfaction on fathers with their partner caring for their infant with bilateral cleft. However, more research is needed in order to explore the perception

that mothers and fathers have of each other in relation to their parental role in the context of CL/P malformation.

Besides, the fact that the impact on family –reported by mothers and fathers- did not differ between parents having an infant with unilateral or bilateral malformation, is in line with other studies in which the size of the cleft defect (unilateral or bilateral) had no relevant influence on any dimension of the Impact On Family Scale (Kramer et al., 2007). It should be noted that, at twelve months, no significant differences were observed between the unilateral and the bilateral cleft groups.

In regard to the **secondary hypothesis** related to exploring the differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the type of diagnosis performed, it has been found that, at four months, the impact on family –reported by both parents- was significantly higher in the prenatal diagnosis group, than in the postnatal diagnosis group. This outcome may be because the prenatal diagnosis may constitute a long-lasting psychological stressor for parents, given the increased latency period between the diagnosis and the treatment availability (Brosig et al., 2007).

Likewise, some studies observed that few parents had not wanted to know about the CL/P malformation during the prenatal period because they thought it would have been more distressing than a postnatal diagnosis (Berggren et al., 2012; Davalbhakta & Hall, 2000; Johansson & Ringsberg, 2004; Nusbaum et al., 2008; Rey-Bellet & Hohlfeld, 2004; Sharp, Strauss, & Lorch, 1992).

Also, our findings are consistent with previous studies. For instance, Kramer et al. (2007) found that prenatal diagnosis of CL/P malformation did not contribute to reduce the impact on family, besides, the social impact was increased in families that were informed before the infant birth. In the same way, it has been seen that parents

of infants with a prenatal diagnosis reported higher levels of psychological distress 6 weeks after the infant birth (Skari et al., 2006) and 6 months after the diagnosis (Brosig et al., 2007), in comparison to parents who received a postnatal diagnosis.

Even when our results indicate a higher impact on family when the diagnosis is prenatal, it is important to consider that the medical team have the obligation to inform the diagnosis of any disease or malformation at the moment of noticing. Consequently, our study emphasizes the importance of providing information about the CL/P malformation when the parents are informed about the diagnosis. As well, it is important to explain about the treatment options to families soon after diagnosis in order to reduce the psychological concern. In this line, psychological support should be offered to parents in order to facilitate their coping strategies.

Additionally, at twelve months, it has been observed that the quality of the parent-infant relationship was significantly higher in the prenatal diagnosis group, in comparison with the postnatal diagnosis group. These results are consistent with a previous study in which it was observed that prenatal diagnosis had a positive impact on the future mother-infant relationship (Baker et al., 2009). Thus, we can hypothesize that the prenatal diagnosis is often seen as advantageous at long-term as it makes it possible for the parents to go through the grieving process and deal with distressing emotions during pregnancy and during the first months after the birth. However, further research is necessary to explore more variables related to the type of diagnosis and the quality of parent-infant relationship at long-term, since the levels of depression or psychological distress may influence the interactive patterns in parents and infants.

In relation to the **secondary hypothesis** related to exploring the differences in infant social withdrawal behavior, psychological impact on parents, and quality of the parent-infant relationship between the

early surgery group and the late surgery group, in the first assessment, we found that impact on family –reported by mothers and fathers- and maternal depressive symptoms were significantly higher in the late surgery group.

Furthermore, in the second evaluation, the impact of the infant's malformation on family –reported by both parents- was significantly higher in the late surgery group, in comparison to the early surgery group. Similarly, paternal psychological distress and depressive symptoms –in mothers and in fathers- were significantly greater in the late surgery group.

These results indicate that the late repair surgery carries greater psychological difficulties for the parents. This is in line with previous studies that suggest that early interventions reduce the esthetical impact of the malformation on parents and family. For example, Galinier et al. (2008) affirmed that early repair lip surgery is essential for the psychological status of the parents. Likewise, Murray et al. (2008) observed that mothers of infants with a late repair surgery were less positively involved and sensitive, and they looked less at their infant. These authors argued that prior to repair surgery, having a disfigured infant may adversely affect maternal mental health. A similar situation could also occur to the parents in our sample.

Some previous studies did not find differences according to the timing of the repair surgery. For example, Slade, Emerson and Freedlander (1999) did not observe evidence to support the idea that early or late repair led to differential levels of anxiety, depressive symptoms or differences in attachment to the infant. However, these authors compared neonatal surgeries vs. 3-month surgery. This contradiction may be due to the fact that in our study the early group includes infants with repair surgery between 0 to 90 days after birth, and the late surgery group comprises infants with a surgery from 91 days and beyond. Additionally, in the qualitative analysis of the data, Slade,

Emerson and Freedlander (1999) found that most mothers preferred their infant to receive an early repair surgery.

It should be noted that, in our results, at four months, it was already evident that the late surgery group showed more difficulties. Interestingly, at twelve months, it was observed more significant differences between the early and the late surgery groups. This may indicate that the timing of the repair surgery had consequences in the short-term, but especially a long-term effect on psychological impact on parents.

4.1. Conclusions

Concerning the general objective, it has been observed a higher psychological impact -in terms of infant social withdrawal behavior, impact of the CL/P malformation on family, paternal psychological distress and depressive symptoms on fathers- in the first evaluation, in comparison with the second assessment. In other words, the psychological impact of the CL/P malformation was greater, on parents and infants, in the first months after the infant birth. This may indicate a long-term adaptation of parents and infants to the medical condition.

Moreover, in both assessments, the level of socially withdrawn infants with CL/P was similar, or even higher, to that observed in the risk population. This outcome emphasized the necessity of evaluating the social withdrawal behavior in infants with CL/P, especially during the first months of life.

The quality of the parent-infant relationship was adaptive in most of the cases, while the impact on the family and the psychological distress on parents were moderate. Besides, depressive symptoms, in our sample, were higher than in general population. However, according to Fonseca, Nazaré, and Canavarro (2012), depressive symptoms can be considered an expected and normative expression of the parental

experience, when there is a diagnosis of a congenital medical condition, given the unexpectedness (Lalor, Begley, & Galavan, 2009) and significance of the loss of a healthy infant (Jones, Statham, & Solomou, 2005).

Interestingly, self-blame was the feeling more reported by both parents in our sample. Accordingly, provide information surrounding the etiology of CL/P and the causal factors is particularly important during the first year after the birth of an infant with CL/P, because this information may reduce the guilt on parents.

Additionally, levels of marital satisfaction of parents having an infant with CL/P were similar to the normal population. This is in line with previous studies that postulate that having a child with illness or disability does not necessarily pose a threat to the parents' marital relationship (Dale et al., 2013). This satisfying marital relationship may act as a protective factor for social development in infants having CL/P and for the quality of the parent-infant relationship.

Furthermore, related to the secondary objectives, our results clearly showed that parents of infants with Cleft Lip and Palate (CLP), reported higher levels of psychological impact, compared to those in the Cleft Lip group (CL). This confirms the greater necessity of psychological support in parents having an infant with CLP, compared to CL infants, during the first year postpartum.

Surprisingly, in the comparison between unilateral and bilateral malformation, fathers of infants with bilateral malformation reported significantly higher marital satisfaction, at four months. However, at twelve months, significant differences were not observed. This would indicate that the laterality of the malformation does not necessary influence the well-being of the parents having an infant with CL/P, specifically in the marital relationship.

When comparing the type of diagnosis groups, prenatal diagnosis group had higher impact on family, at four months evaluation, in comparison with the postnatal diagnosis group. This leads to consider that parents who learn the diagnosis of CL/P in the prenatal period may need psychological support throughout the pregnancy in order to avoid the risk of misinformation, to reduce the psychological impact, and to help them to recover from the loss of the imagined healthy infant.

At twelve months, the quality of parent-infant relationship was higher when the diagnosis was given prenatally. This may indicate that, in the long-term, a prenatal diagnosis of CL/P may be a protector factor for the parent-infant interaction. However, further research is necessary in order to determine other variables that may be related to the quality of the parent-infant relationship and the time of the diagnosis.

One of the most important contributions of our study was that an early lip repair surgery (performed during the first 90 days of infant's life) significantly reduces the psychological impact of the CL/P malformation on parents, and contributes to preserve the parental psychological state, during the first year after the infant's birth. These findings provide valid and necessary information to facilitate the recognition and the approval of a surgical protocol –by the medical community- that takes into account the psychological perspective of the infant and their parents.

In summary, we observed that parents and infants, in our sample, showed a good adaptation to the CL/P malformation, during the first year postpartum, since the level of infant social withdrawal and psychological impact on parents were lower in the second evaluation, in comparison to the first one. Moreover, this adaptation is better when the infant has a simpler malformation (Cleft Lip) versus a more severe malformation (Cleft Lip and Palate). The prenatal diagnosis showed to improve, in the long term, the quality of parent-infant relationship. In addition, the repair surgery performed during the first 90 days after

birth, helped to preserve the psychological wellbeing of the parents. These indicators allow for the early detection of families and infants at higher risk of psychological distress, and provide guidelines for clinical practices.

4.2. Practical implications

Our results suggest that having an infant with CL/P malformation is a major psychological challenge for the infants and their parents. We found some indicators that facilitate the identification of families with a major risk of psychological distress. We observed that parents having an infant with a more severe malformation (CLP) need a major emotional support, in comparison with the CL group.

The prenatal diagnosis showed to improve, in the long term, the quality of parent-infant relationship. Accordingly, parents who have known the diagnosis postnatally require more psychological support in order to improve their interactional patterns with their infant with CL/P. In addition, it was found that an early lip repair surgery has more benefits for parents in terms of less psychological impact, in comparison to a late surgery. Medical community should be informed about all these indicators in order to refer families with major risk of psychological distress, and to prioritize repair surgeries during the first three months postpartum.

Moreover, our results revealed that the evaluation of infant social withdrawal behavior –during the first months postpartum- is essential in the context of CL/P malformation in order to detect signs of psychological distress and interactional difficulties. However, psychologists should take into account that social withdrawal behavior is one of the signs that infants can manifest in the context of psychological distress. Thus, it is important to be alert to other expressions of emotional suffering, such as sleep and feeding disorders.

Although symptoms of psychological impact on infants and parents may be understood as the result of a normative process of individual adjustment to the stress-inducing event (Boss, 2002) –in this case the CL/P malformation- it is indispensable to evaluate all these symptoms in a comprehensive approach, taking into account the perspective of the infants and both mothers and fathers. Besides, parents and infants should receive psychological support since the diagnosis announcement and through all the prenatal and the early postnatal period.

In this line, previous studies have found that a higher proportion of families deplored the lack of knowledge and sensitivity of care teams at birth (e.g., Rey-Bellet & Hohlfeld, 2004). Hence, basic information should be provided at the moment of the diagnosis confirmation, followed by progressively more detailed technical discussions over the remaining pregnancy in order to reduce parental concern. In this way, Rey-Bellet and Hohlfeld (2004) insisted that it is essential for medical teams and psychologists to help parents to *invest* in their infant –who is different than the expected and hoped for- but also to help them become their infant's care providers, by listening to them.

For instance, psychoeducation about the CL/P malformation should be offer to parents to reduce the risk of misinformation. The etiology of the CL/P malformation, its implications on the patient, the different treatments, and the repair surgical interventions should be explained to parents to decrease fear and guilt. Furthermore, it is important to give parents the opportunity to express their emotions, perceptions and doubts about the CL/P malformation. Parents may be also benefit from decision-making strategies, problem solving training, coping strategies, and emotional expression strategies. In addition, psychologists and the medical team can encourage parents to focus on their infant's positive attributes, rather than on physical and functional characteristics (Eiserman, 2001; Rumsey & Harcourt, 2005).

All this highlights the importance of including a psychologist as part of the interdisciplinary team caring CL/P families in order to offer specialized and more targeted psychological support to parents and infants with higher difficulties. Specialized interventions may benefit parents and infants coping with CL/P malformation to facilitate the adaptation to transition to parenthood and to the several stressors associated to the CL/P malformation.

4.3. Strengths and limitations

A number of limitations of this research must be acknowledged. First, a larger and similar number of infants included in all groups is required in order to do comparisons among them, and thus, be able to achieve more specific outcomes according to the type and laterality of the malformation, the type of diagnosis and the timing of the repair surgery. However, due to the medical condition of this sample, the recruitment of participants entails greater difficulties compared with general population studies.

Second, most of the parents in our sample were in a long-term relationship and were employed at the time of the study. As a result, those with diverse ethnic backgrounds, those reporting a different marital status or less traditional family structure, and those living in lower socioeconomic conditions were not represented in this research. Similar bias was identified in other studies (Costigan & Cox, 2011; Stock & Rumsey, 2015) in which fathers with less education, less traditional family structures, and less optimal environments were less likely to participate in family research. Accordingly, it is important to take into account this characteristics of the sample when interpreting the results of the current study.

Third, another possible limitation comes from our decision to base parental psychological impact on self-report questionnaires, which may biased by social desirability. Moreover, there was some missing data

that it was not possible to recover, thus future investigations should pay major attention during the data collection. In addition, there was no record of the reasons why some families (15.17%) did not return to the second evaluation. However, as the results of the IOFS indicated, families find it exhausting to go to the hospital appointments, so this may be the reason why some of them did not come to the twelve months assessment. A closer follow-up by the medical team to the families may reduce the percentage of missing sample in the second evaluation.

Given that the first assessment of parents and infants was carried out at four months postpartum, it is important that future research examine the psychological impact on parents and infants in the context of CL/P malformation from birth to three months, in order to design and to implement specific and targeted interventions, as soon as possible, that can effectively prevent and reduce the psychological burden on families having an infant with CL/P.

Further studies should explore psychological impact on unaffected siblings in order to have a more comprehensive understanding of how families as a unit are impacted by having a member with CL/P. Also, future research should be focused more on resilience and coping strategies in families having an infant with CL/P in order to promote more adaptive strategies in parents.

The limitations of our study are inherent in research in clinical context with pre-post evaluations. The data that come from this type of clinical sample are considered especially relevant due to the difficulty to obtain them and to assure their quality.

One of the strengths of the current research was the adoption of a psychological comprehensive approach to parental and infant adjustment to CL/P malformation. This is the first study that provides information about social withdrawal behavior in infants with CL/P

during the first year postpartum. Besides, families in our study came from different ethnics, which may be considered representative of the French population. In this line, in terms of family incomes, these families were within the average.

It should be noted that in all assessment instruments, the values of Cronbach's alpha indicates that internal consistency was good-acceptable, which guarantees their use in infants having CL/P and their families. This may be considered another contribution of our study.

Moreover, we explored not only the parents' perspective, but also the infant's psychological distress, through the infant social withdrawal, and the quality of the parent-infant relationship. In addition, exploring the psychological impact on both parents, rather than just mothers, was another strength of our study. Taking into account the perspective of fathers provides an overall perspective of the adaptation of the parental couple to face the CL/P malformation of their infant. This approach allows to obtain a more complete profile of the psychological impact of the CL/P malformation on parents and infants, and provides indicators to identify families with a major risk of psychological distress.

The longitudinal design of the study allowed to explore the evolution of infant social withdrawal behavior, the quality of parent-infant relationship and the psychological impact of the CL/P malformation on parents. In addition, this longitudinal design endorsed the possibility to explore these variables in infants having an early or late repair surgery. Longitudinal studies of repeated measures are highly valued by medical and psychological research because of their contribution to good clinical practice.

Finally, the results of our research allow a psychological approach of the Cleft Lip and Palate (CL/P) malformation from an evidence-based practice perspective (EBP), providing data that have a direct

implication in clinical practice, and that facilitate clinical decision-making to improve the follow-up and the treatment of infants with CL/P and their parents.

5. REFERENCES

Abbott, B. D. (2010). The etiology of cleft palate: a 50-year search for mechanistic and molecular understanding. *Birth Defects Research. Part B Developmental and Reproductive Toxicology, 89,* 266–274.

Albuquerque, S., Pereira, M., Fonseca, A., & Canavarro, C. (2012). Family impact and individual adjustment of parents of children with a diagnosis of congenital anomaly: the influence of the child's determinants. *Revista de Psiquiatría Clínica*, *39* (4), 136-141.

Alderfer, M. A., & Kazak, A. E. (2006). Family issues when a child is on treatment for cancer. In R. T. Brown (Ed.), *Comprehensive handbook of childhood cancer and sickle cell disease: A biopsychosocial approach* (p. 53–74). New York: Oxford University Press.

Almesned, S., Al-Akhfash, A., & Al Mesned, A. (2013). Social impact on families of children with complex congenital heart disease. *Annals of Saudi Medicine, 33 (2),* 140-143.

American Cleft Palate Craniofacial Association. (2009). *Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies-Revision.* Available in: http://www.acpa-cpf.org/uploads/site/Parameters_Rev_2009.pdf

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th Ed.). Washington, DC: Author.

Anders, T.F. (1989). Clinical syndromes, relationship disturbances, and their assessment. In A.J. Sameroff & R.N. Emde (Eds.), *Relationship disturbances in early childhood* (p. 125–144). New York: Basic Books.

Anderson, K. N. (Ed.), (1998). *Mosby's medical, nursing and allied health dictionary* (5th Ed.). St Louis: Mosby.

Antoine, P., Christophe, V., & Nandrino, J.L. (2008). Échelle d'ajustement dyadique: intérêts cliniques d'une révision et validation d'une version abrégée. *L'Encéphale, 34*, 38-46.

Apter-Levy, Y., Feldman, M., Vakart, A., Ebstein, R. P., & Feldman, R. (2013). Impact of maternal depression across the first 6 years of life on the child's mental health, social engagement, and empathy: The moderating role of oxytocin. *American Journal of Psychiatry,* 170, 1161–1168. doi:10.1176/appi.ajp.2013.12121597

Bagner, D. M., Pettit, J. W., Lewinsohn, P. M., & Seeley, J. R. (2010). Effect of maternal depression on child behavior: a sensitive period? *Journal of the American Academy of Child and Adolescent Psychiatry*, *49* (7), 699-707.

Bakeman, R., & Brown, J.V. (1980). Early interaction: Consequences for social and mental development at three years. *Child Development, 51*, 437–447.

Baker, S.R., Owens, J., Stern, M., & Willmot, D. (2009). Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *The Cleft Palate-Craniofacial Journal, 46,* 229–36.

Barden, R.C., Ford, M.E., Jensen, A.G., Rogers-Salyer, M., & Salyer, K.E. (1989). Effects of craniofacial deformity in infancy on the quality of mother-infant interactions. *Child Development, 60*, 819–824.

Baumann, S.L., & Braddick, M. (1999) Out of their element: fathers of children who are "not the same". *Journal of Pediatric Nursery, 14* (6), 369–378.

Bayley, N. (1993) *Bayley Scales of Infant Development* (2nd Ed.). San Antonio, TX: The Psychological Corporation.

Beebe, B. (1982). Micro-timing in mother–infant communication. In M. Key (Ed.), *Nonverbal communication today: Current research* (p. 169–195). New York: Mouton.

Beebe, B., Lachmann, F., & Jaffe, J. (1997). Mother-infant structures and pre-symbolic self and object representations. *Psychoanalytic Dialogues, 2,* 133–182.

Bellerose C. (1995). *Et la santé, ça va en 1992–1993? Rapport de l'enquête sociale et de santé 1992–1993.* Montreal: Santé Québec.

Benson, B. A., & Gross, A. M. (1989). The effect of a congenitally handicapped child upon the marital dyad: A review of the literature. *Clinical Psychology Review, 9,* 747–758.

Berggren, H., Hansson, E., Uvemarck, A., Svensson, H. & Becker, M. (2012). Prenatal compared with postnatal cleft diagnosis: What do the parents think? *Journal of Plastic Surgery and Hand Surgery, 46,* 235-241.

Berk, N.W., Cooper, M.E., Liu, Y.E., & Marazita, M.L. (2001). Social anxiety in Chinese adults with oral-facial clefts. *Cleft Palate Craniofacial Journal, 38* (2), 126–33.

Bevilacqua, F., Palatta, S., Mirante, N., Cuttini, M., Seganti, G., Dotta, A. & Piersigilli, F. (2013). Birth of a child with congenital heart disease: emotional reactions of mothers and fathers according to time of diagnosis. *Journal of Maternal-Fetal and Neonatal Medicine*, *26*, 1249–1253

Bodenmann, G. (1997). Dyadic coping: A systemic-transactional view of stress and coping among couples: Theory and empirical findings. *Revue Européenne de Psychologie Appliquée*, *47*, 137–140.

Boudas, R., Jégu, J., Grollemund, B., Quentel, E.m Danion-Grilliat, A., & Velten, M. (2013). Cross-cultural French adaptation and validation of the Impact On Family Scale (IOFS). *Health and Quality of life Outcomes, 11*: 67. Available in http://hqlo.biomedcentral.com/articles/10.1186/1477-7525-11-67

Boss, P. (2002). *Family Stress Management: A Contextual Approach*. 2nd ed. London, UK: SAGE.

Bowman, M.L. (1990). Coping effort and marital satisfaction: measuring marital coping and its correlates. *Journal of Marriage and the Family*, *52* (2), 463-474.

Braarud, H., Slinning, K., Moe, V., Smith, L., Vanned, U., Guedeney, A., & Heimann, M. (2013). Relation between social withdrawal symptoms in full-term and premature infants and depressive symptoms in mother: a longitudinal study. *Infant Mental Health Journal, 34* (6), 532–541.

Bradbury, E., & Hewison, J. (1994). Early parental adjustment to visible congenital disfigurement. *Child: Care, Health and Development, 20*, 251–266.

Brand, S., Blechschmidt, A., Müller, A., Sader, R., Schwenzer-Zimmerer, K., Zeilhofer, H.F., & Holsboer-Trachsler, E. (2009). Psychosocial functioning and sleep patterns in children and adolescents with cleft lip and palate (CLP) compared with healthy controls. *Cleft Palate Craniofacial Journal, 46* (2), 124–35.

Brazelton, B. (1973). *Neonatal Behavioral Assessment Scale.* Philadelphia: Lippincott.

Brazelton, B., & Cramer, B. G. (1990). *The earliest relationship*. New York: Addison-Wesley.

Brazelton, T.B., Koslowski, B., & Main, M. (1974). The origins of reciprocity in early mother–infant interaction. In M. Lewis & L.A. Rosenblum (Eds.), *The effects of the infant on its caregiver* (p. 49–75). New York: Wiley.

Brehaut, J.C., Kohen, D., Garner, R., Miller, A., Lachm L., Klassen, A.F., & Rosenbaum, P. (2009). Health among caregivers of children with health problems: Finding from a Canadian population-based study. *American Journal of Public Health, 99* (7), 1254-1262.

Broder, H.L., Richman, L.C., & Matheson, P.B. (1998). Learning disability, school achievement, and grade retention among children with cleft: a two-center study. *Cleft Palate Craniofacial Journal, 35,* 127–131.

Brosig, C.L., Whitstone, B.N., Frommelt, M.A., Frisbee. S. J., & Leuthner, S. R. (2007). Psychological distress in parents of children with severe congenital heart disease: the impact of prenatal versus postnatal diagnosis. *Journal of Perinatology, 27,* 687-692. Available in: https://www.nature.com/jp/journal/v27/n11/full/7211807a.html

Brown, R.T., Kaslow, N.J., Hazzard, A.P., Madan-Swain, A., Sexson, S.B., Lambert, R., & Baldwin, K. (1992). Psychiatric and family functioning in children with leukemia and their parents. *Journal of the American Academy of Child and Adolescent Psychiatry, 31* (3), 495–502. doi: 10.1097/00004583-199205000-00017

Bull, R., & Rumsey, N. (1988). The social psychology of facial disfigurement. *Journal of Cross-Cultural Psychology*, *17*, 99–108.

Burtchen, N., Álvarez-Segura, M., Mendelsohn, A., Dreyer, B., Castellanos, F., Catapano, P., & Guedeney, A. (2013). Screening for sustained social withdrawal behaviors in six-month-old infants during pediatric primary care visits: results from an at-risk Latino immigrant simple with high rates of maternal major depressive disorder. *Infant Mental Health Journal, 34* (6), 542-552.

Cambonie, G., Muller, J.B., Ehlinger, V., Guedeney, A., Lebeaux, C., Kaminski, M., Alberge, C., Denizot, S., Ancel, P.Y., Arnaud, C.

(2017). Mother-infant interaction assessment at discharge and at 6 months in a French cohort of infants born very preterm: the OLIMPE study. (Submitted).

Canfield, M. A., Honein, M. A., Yuskiv, N., Xing, J., Mai, C. T., Collins, J. S., Kirby, R. S., the National Birth Defects Prevention Network. (2006). National estimates and race/ethnic-specific variation of selected birth defects in the United States, 1999-2001. *Birth Defects Research, Part A, 76*, 747–756.

Chapman, K. L., Hardin-Jones, M., Schulte, J., & Halter, K. (2001). Vocal development of 9-month-old babies with cleft palate. *Journal of Speech, Language, and Hearing Research*, *44,* 1268-1283.

Cheung, L.K., Loh, J.S., & Ho, S.M. (2007) Psychological profile of Chinese with cleft lip and palate deformities. *Cleft Palate Craniofacial Journal, 44*, 79–86.

Chetpakdeechit, W., Hallberg, U., Hagberg, C., & Mohlin, B. (2009). Social life aspects of young adults with cleft lip and palate: grounded theory approach. *Acta Odontologica Scandinavica, 67*, 122–128.

Christ, J.E., & Meininger, M. G. (1981). Ultrasound diagnosis of cleft lip and cleft palate before birth. *Plastic and Reconstructive Surgery, 68*, 854-859.

Christensen, A., Atkins, D.C., Berns, S., Wheeler, J., Baucom, D.H., & Simpson, L.E. (2004). Traditional versus integrative behavioral couple therapy for significantly and chronically distressed married couples. *Journal of Consulting and Clinical Psychology, 72*, 176–191.

Christensen, K., Juel, K., Herskind, A.M., & Murray, J.C. (2004). Long term follow up study of survival associated with cleft lip and palate at birth. *BMJ, 28,* 1405–1408.

Christensen, K., & Mortensen, P.B. (2002). Facial clefting and psychiatric diseases: a follow-up of the Danish 1936–1987 Facial Cleft cohort. *Cleft Palate Craniofacial Journal*, *39* (4), 392–396.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd Ed.). Hillsdale, N.J.: Lawrence Erlbaum Associates.

Cohn, J., & Tronick, E.Z. (1983). Three-month-old infants' reaction to simulated maternal depression. *Child Development*, 54, 185–193.

Cohn, J., & Tronick, E. Z. (1987). Mother-infant face-to-face interaction: the sequence of dyadic states at 3-6 and 9 months. *Developmental Psychology, 23*, 68-77.

Cohn, J., & Tronick, E.Z. (1989). Specificity of infants' response to mothers' affective behavior. *Journal of the American Academy of Child and Adolescent Psychiatry, 28*, 242–248.

Collett, B. R., & Speltz, M. L. (2007). A developmental approach to mental health for children and adolescents with orofacial clefts. *Orthodontics & Craniofacial Research, 10*, 138 –148.

Conway, J.C., Taub, P.J., Kling, R. Oberoi, K. Doucette, J. & Jabs, E.W. (2015). Ten-year experience of more than 35,000 orofacial clefts in Africa. *BioMed Central Pediatrics. 15* (8), 1-9.

Corbo Rodríguez, M.T., & Torres E.M. (2001). Labio y paladar fisurados, aspectos generales que se deben conocer en la atención primaria de salud. *Revista Cubana de Medicina General Integral, 4*, 379-385.

Costigan, C.L., & Cox, M.J. (2001). Fathers' participation in family research: is there a self-selection bias? *Journal of Family Psychology, 15*, 706–720.

Costa, R., & Figuereido, B. (2012). Infant's behavioral and physiological profile and mother–infant interaction. *International Journal of Behavioral Development*, 36 (3), 205–214.

Costa, R., & Figueiredo, B. (2013). The Alarm distress Baby Scale in a longitudinal Portuguese study reanalyzed with attachment data. *Infant Mental Health Journal*, *34* (6), 553-561.

Cowan, C. P., Cowan, P. A., Heming, G., & Miller, N. (1991). Becoming a family: Marriage, parenting and child development. In P. A. Cowan & M. Hetherington (Eds.), *Family transitions* (p. 79-110). Hillsdale, NJ: Erlbaum.

Cowan, C. & Cowan, P. (1992). *When partners become parents: The big life changes for couples.* New York: Basic Books.

Cox, J.L., Chapman, G., Murray, D., & Jones, P. (1996). Validation of the Edinburgh postnatal depression scale (EPDS) in non-postnatal women. *Journal of Affective Disorders*, *39*, 185-189.

Cox, J.L., Holden, J.M., & Sagovsky, R. (1987). Detection of postnatal depression: Development of the 10-item. Edinburgh Postnatal Depression Scale. *British Journal of Psychiatry*, *150*, 782-786.

Crane, D.R., Allgood, S.M., Larson, J.H., & Griffin, W. (1990). Assessing marital quality with distressed and non-distressed couples: A comparison and equivalency table for three frequently used measures. *Journal of Marriage and the Family, 52*, 87–93.

Dale, M. T.G., Solberg, O., Holmstrom, H., Landolt, M. A., Eskedal, L., & Vollrath, E. (2013). Relationship satisfaction among mothers of children with congenital heart defects: A prospective case-cohort study. *Journal of Pediatric Psychology*, *38* (8), 915-926.

Davalbhakta, A., & Hall, P.N. (2000). The impact of antenatal diagnosis on the effectiveness and timing of counselling for cleft lip and palate. *British Journal of Plastic Surgery*, *53,* 298–301.

Davies, P., Cummings, E., & Winter, M. (2004). Pathways between profiles of family functioning, child security in the interparental subsystem, and child psychological problems. *Development and Psychopathology*, *16,* 525-550.

De Pascalis, L., Kkeli, N., Chakrabarti, B., Dalton, L., Vaillancourt, K., Rayson, H., Bicknell, S., Goodacre, T., Cooper, P., Stein, A. & Murray, L. (2017). Maternal gaze to the infant face: Effects of infant age and facial configuration during mother-infant engagement in the first nine weeks. *Infant Behavior and Development, 46,* 91-99.

De Rosa, E., Curró, V., Wendland, J., Maulucci, S., Maulucci, M. L., & De Giovanni, L. (2010). Propriétés psychométriques de l'échelle Alarme Détresse Bébé (ADBB) appliquée à 81 enfants italiens. Dépression postnatale, maladie somatique maternelle et retrait chez l'enfant. *Devenir, 3* (22), 209-223.

De Sousa, A., Devare, S., & Ghanshani, J. (2009). Psychological issues in cleft lip and palate. *Journal of Indian Association of Pediatric Surgeons, 14* (2), 55-58. doi: 10.4103/0971-9261.55152

Dehn, L.B., Korn-MErker, E., Pfafflin, M., Ravens-Sieberer, U., & May, T. W. (2014). The Impact on Family Scale: Psychometric analysis of long and short forms in parents of children with epilepsy. *Epilepsy and Behavior*, *32,* 21-26.

Deroo, L. A. & Wilcox, A. J. (2008). First-trimester maternal alcohol consumption and the risk of infant oral clefts in Norway: a population-based case–control study. *American Journal of Epidemiology, 168,* 638–646.

Despars, J, Peter, C, Borghini, A, Pierrehumbert, B., Habersaat, S., Müller-Nix, C., ansermet, F. & Hohlfeld, J. (2011). Impact of a cleft lip and/or palate on maternal stress and attachment representations. *Cleft Palate Craniofacial Journal, 48*, 419–424.

Dissaux, C., Bodin, F., Grollemund, B., Picard, A., Vazquez, M. P., Morand, B., James, I., Kauffman, I. & Bruant-Rodier, C. (20159. Evaluation of 5-year-old children with complete cleft lip and palate: Multicenter study. Part 1: Lip and nose aesthetic results. *Journal of Cranio-Maxillo-Facial Surgery*, *43,* 2085-2092. http://dx.doi.org/10.1016/j.jcms.2015.08.020

Dixon, M.J., Marazita, M. L., Beaty, T.H., & Murray, J. (2011). Cleft lip and palate: understanding genetic and environmental influences. *Nature Reviews Genetics*, *12,* 167-178. doi: 10.1038/nrg2933

Doherty, N., McCusker, C. G., Molly, B., Mulholland, C., Rooney, N., Craig, B., Sands, A., Stewart, M., & Casey, F. (2009). Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease. *Journal of Reproductive and Infant Psychology, 27* (4), 390-400.

Dölger-Hafner, M., Bartsch, A., Trimbach, G., Zobe,l I., & Witt, E. (1997). Parental reactions following the birth of a cleft child. *Journal of Orofacial Orthopedics, 58,* 124–133.

Dollberg, D., Feldman, R., Keren, M., & Guedeney, A. (2006). Sustained withdrawal behavior in clinic referred and non-referred infants. *Infant Mental Health Journal, 27* (3), 292–309.

Downey, G., & Coyne, J.C. (1990). Children of depressed parents: an integrative review. *Psychological Bulletin*, *108*, 50–76.

Drotar, D., Baskiewicz, A., Irvin, N., Kennell, J., Kennell, J., & Klaus, M. (1975). The adaptation of parents to the birth of an infant with a congenital malformation: A hypothetical model. *Pediatrics, 56,* 710–717.

Eiserman, W. (2001). Unique outcomes and positive contributions associated with facial difference: expanding research and practice. *Cleft Palate Craniofacial Journal*, *38,* 236–244.

Emde, R.N., & Sameroff, A.J. (1989). Understanding early relationship disturbances. In A.J. Sameroff & R.N. Emde (Eds.), *Relationship disturbances in early childhood* (p. 3–16). New York: Basic Books.

Endriga, M.C., & Speltz, M.L. (1997). Face-to-face interaction between infants with orofacial clefts and their mothers. *Journal of Pediatric Psychology*, 22, 439–453.

Engel, G.L., & Reichsman, F. (1956). Spontaneous and experimentally induced depression in an infant with gastric fistula: a contribution to the problem of depression. *Journal of the American Psychoanalytical Association, 4,* 428–452.

Engel, G.L., & Reichsman, F. (1979). Monica: a 25 years follow-up longitudinal study of the conse- quences of trauma in infancy. *Journal of the American Psychoanalytical Association*, *27,* 107–126.

Engel, G.L., & Schmale, A.H. (1972). Conservation withdrawal: a primary regulatory process for organismic homeostasis. In D. Hill (Ed), *Physiology, emotion and psychosomatic illness* (p. 57–85). Amsterdam: Elsevier Excerpta Medica.

Espié, E., Ouss, L., Gaboulaud, V., Candilis, D., Ahmed, K., Cohuet, S., Baubet, T., Grais, R., & Moro, M. R. (2011). Against the Odds: Psychomotor Development of Children under 2 years in a Sudanese orphanage. *Journal of Tropical Pediatrics, 57* (6), 412-417.

Evans, J., Melotti, R., Heron, J., Ramchandani, P., Wiles, N., Murray, L., & Stein, A. (2012). The timing of maternal depressive symptoms and child cognitive development: A longitudinal study. *Journal of Child Psychology and Psychiatry, 53* (6), 632-640.

Facuri-Lopes, S., Ricas, J., & Cotta-Mancini, M. (2008) Evaluation of the psychometric properties of the Alarm Distress Baby Scale among 122 Brazilian children. *Infant Mental Health Journal 29* (2), 153–173.

Farroni, T., Csibra, G., Simion, F., & Johnson, M. H. (2002). Eye contact detection in humans from birth. *Proceeding of the National Academy of Sciences*, *99* (14), 9602-9605.

Favez, N., Frascarolo, F., Keren, M., & Fivaz-Depeursinge, E. (2009). Principles of Family Therapy in Infancy. In C. H. Zeanah (Ed.),

Handbook of infant mental health (p.468-484). New York: The Guildford Press.

Fearnley Shapiro, A., Gottman, J. M., & Carrière, S. (2000). The baby and the marriage: Identifying factors that buffer against decline in marital satisfaction after the first baby arrives. *Journal of Family Psychology, 14*, 59–70.

Feldman, R. (2007). Parent–infant synchrony and the construction of shared timing; physiological precursors, developmental outcomes, and risk conditions. *Journal of Child Psychology and Psychiatry*, *48,* 329–354.

Feldman, R., & Eidelman, A.I. (2003). Direct and indirect effects of maternal milk on the neurobehavioral and cognitive development of premature infants. *Developmental Psychobiology*, *43*, 1–12.

Field, T. (1977). Effects of early separation, interactive deficits, and experimental manipulations on mother-infant face-to-face interaction. *Child Development*, *48,* 763–771.

Field, T. (1987). Affective and interactive disturbances in infants. In J. Osofsky (Ed.), *Handbook of infant development* (p. 972–1005). New York: Wiley.

Field, T.M., Cohen, D., García, R., & Greenberg, R. (1984). Mother-stranger face discrimination by the newborn. *Infant Behavior and Development*, *7*, 19-25.

Field, T.M., & Vega-Lahr, N. (1984). Early interactions between infants with craniofacial anomalies and their mothers. *Infant Behavior and Development, 7*, 527–530.

Fiese, B., Poehlmann, J., Irwin, M., Gordon, M., & Curry-Bleggi, E. (2001). A pediatric screening instrument to detect problematic infant-parent interactions: Initial reliability and validity in a sample of high-and low-risk infants. *Infant Mental Health Journal, 22* (4), 463-478.

Fonseca, A., Nazaré, B. & Canavarro, M.C. (2012). Parental psychological distress and quality of life after a prenatal or postnatal diagnosis of congenital anomaly: A controlled comparison study with parents of healthy infants. *Disability and Health Journal, 5*, 67-74.

Fonseca, A., Nazaré, B. & Canavarro, M.C. (2013). Parental psychological distress and confidence after an infant's birth: The

role of attachment representations in parents of infants with congenital anomalies and parents of healthy infants. *Journal of Clinical Psychology in Medical Settings, 20,* 143-155.

Fonseca, A., Nazaré, B. & Canavarro, M.C. (2015). Parenting an infant with a congenital anomaly: How are perceived burden and perceived personal benefits related to parenting stress? *Journal of Clinical Psychology in Medical Settings*, *22*:64. doi: 10.1007/s10880-015-9418-7

Fogel, A. (1993). *Developing through relationships*. Chicago: University of Chicago Press.

Fox, N. A. (2004). Temperament and early experience form social behavior. *Annals of the New York Academy of Sciences*, *1038,* 171–178.

Fox, G. L., Nordquist, V. M., Billen, R. M., & Savoca, E. F. (2015). Father involvement and early intervention: Effects of empowerment and father role identity. *Family Relations*, *64*, 461–475. http://dx.doi.org/10.1111/fare.12156

Franck, L.S., & Spencer, C. (2003). Parent visiting and participation in infant caregiving activities in a neonatal unit. *Birth, 30,* 31–5.

Fraiberg, S. (1982). Pathological defenses in infancy. *Psychoanalytical Quarterly, 4*, 612–635.

Fraser, F. C. (1955). Thoughts on the etiology of clefts of the palate and lip. *Acta Genetica et Statistica Medica, 5*, 358–369.

Frías-Navarro, D. (Ed.). (2011). *Técnica estadística y diseño de la investigación.* Valencia: Palmero Ediciones.

Galinier, P., Salazard, B., Deberail, A., Vitkovitch, F., Caovan, C., Chausseray, G., Acar, P., Sami, K., Guitard, J. & Smail, N. (2008). Neonatal repair of cleft lip: a decision-making protocol. *Journal of Pediatric Surgery, 43*, 662-667.

Garner, A.S., Shonkoff, J.P., Siegel, B.S., Dobbins, M.I., Earls, M.F., McGuinn, L., Pascoe, J., & Wood, D.L. (2012). Early childhood adversity, toxic stress, and the role of the pediatrician: Translating developmental science into lifelong health. *Pediatrics*, *129* (1), e224–e231. doi: 10.1542/peds.2011-2662

Gauvain-Piquard, A., Rodary, Ch., Rezvani, A., & Lemerle, J. (1987). Pain in children aged 2–6 years: a new observational rating scale elaborated in a pediatric oncology unit. *Pain, 31,* 177–188.

Gauvain-Piquard, A., Rodary, C., Rezvani, A., & Serbouti, S. (1999). The development of the DEGR: A scale to assess pain in young children with cancer. *European Journal of Pain, 3*, 165–176.

Gavin, N.I., Gaynes, B.N., Lohr, K.N., Meltzer-Brody, S., Gartlehner, G., & Swinson, T. (2005). Perinatal depression: A systematic review of prevalence and incidence. *Obstetrics & Gynecology*, *106,* 1071–1083.

Gaynes, B.N., Gavin, N., Meltaer-Brody, S., Lohr, K.N., Swinson, T., Gartlehner, G., et al. (2005). *Perinatal depression: Prevalence, screening accuracy, and screening outcomes.* Rockville, MD: Agency for Healthcare Research and Quality.

George, D., & Mallery, P. (2003). *SPSS for Windows step by step: A simple guide and reference. 11.0 update* (4th Ed.). Boston: Allyn & Bacon.

George, D., & Mallery, M. (2010). *SPSS for Windows step by step: A simple guide and reference*, *17.0 update* (10a Ed.) Boston: Pearson.

Gianino, A., & Tronick, E. Z. (1988). The mutual regulation model: The infant's self and interactive regulation and coping and defense capacities. In T. Field, P. McCabe, & N. Schneiderman (Eds.), *Stress and coping* (p. 47-68). HiUsdale, NJ: Erlbaum.

Glascoe, F.P., & Macias, M. (2003). Implementing the AAP’s new policy on developmental and behavioral screening. *Contemporary Pediatrics*, 20(4), 85–102.

Goldberg, S., Brachfeld, S., & DiVitto, B. (1980). Feedings, fussing, and playing: Parent-infant interaction in the first year as a function of prematurity and prenatal problems. In T. Field, S. Goldberg, D. Stern, & A. Sostek (Eds.), *High-risk infants and children: adult and peer interactions* (p. 133–153). New York: Academic.

Goodacre, T., Hentges, F., Moss, T., Short, V., & Murray, L. (2004). Does repairing a cleft lip neonatally have any effect on the attractiveness of the subsequent repair? *Cleft Palate-Craniofacial Journal, 41,* 603–608.

Goodacre, T., & Swan, M. C. (2008). Cleft lip and palate: current management. *Pediatrics and Child Health Journal, 18*:6. doi: http://dx.doi.org/10.1016/j.paed.2008.03.008

Goodman, J. H. (2004). Paternal postpartum depression, its relationship to maternal postpartum depression, and implications for family health. *Journal of Advanced Nursing*, *45*, 26-35.

Goodman, S.H., Brogan, D., Lynch, M.E., & Fielding, B. (1993). Social and emotional competence in children of depressed mothers. *Child Development*, *64*, 516–531.

Gray, D. E. (2003). Gender and coping: The parents of children with high functioning autism. *Social Science and Medicine*, *56,* 631–642.

Greenspan, S. (1992). *Infancy and early childhood: The practice of clinical assessment and intervention with emotional and developmental challenges*. Madison: International Universities Press.

Greenspan, S. (2000). *Building healthy minds: The six experiences that create intelligence and emotional growth in babies and young children*. New York: Da Capo Press.

Greenspan, S., & Wieder, S. (1993). Regulatory disorders. In C.H. Zeanah (Ed.), *Handbook of infant mental health* (p. 280–290). New York: Guilford Press.

Gressier, F., Tabat-Bouher, M., Cazas, O., & Hardy, P. (2015). Dépression paternelle du post-partum: revue de la littérature. *La Presse Médicale, 44*, 418-424.

Grollemund B, Danion A, Smaniotto A, Gruillot E. (2011). Visages fendus: réflexions sur la prévention normative des malformations faciales. *Cahiers Philosophiques*, *125,* 30–46. Available in: https://www.cairn.info/revue-cahiers-philosophiques-2011-2-page-30.htm

Grollemund, B., Galliani, E., Soupre, V., Vazquez, M.P., Guedeney, A., & Danion-Grilliat, A. (2010). L'impact des fentes labioplatines sur les relations parent-enfant. *Archives de Pédiatrie*, *17,* 1380-1385. doi:10.1016/j.arcped.2010.06.026

Grollemund, B., Guedeney, A., Vazquez, M.P., Picard, A., Soupre, V., Pellerin, P., Simon, E., Velten, M., Dissauxm C., Kauffmann, I., Bruant-Rodier, C., & Danion-Grilliat, A. (2012a). Relational

development in children with cleft lip and palate: influence of the waiting period prior to the first surgical intervention and parental psychologicall perceptions of the abnormality. *BMC Pediatrics*, *12,* 65. doi: 10.1186/1471-2431-12-65

Grollemund, B., Barrière, M., Guedeney, A., Danion-Grilliat, A. & Groupe CLIP Hôpital Trousseau Paris. (2012b). Fentes labiopalatines : une transmission difficile. *Neuropsychiatrie de l'enfance et de l'adolescence, 60,* 297-301. doi: 10.1016/j.neurenf.2012.02.006

Guedeney, A. (1997). From early withdrawal reaction to infant depression: A baby alone does exist. *Infant Mental Health Journal*, *18,* 339–349.

Guedeney, A. (2015). *Alarm Distress Baby Scale: A recent manual for use.* Available in: http://www.adbb.net/gb-formation.html

Guedeney, A., & Fermanian, J. (2001). A validity and reliability study of assessment and screening for sustained withdrawal reaction in infancy: The Alarm Distress Baby Scale. *Infant Mental Health Journal, 22,* 559–575.

Guedeney, A., Forhan, A. Larroque B., Agostini, M., Pingault, J. B., Heude, B., & the EDEN mother-child cohort study group. (2016). Social withdrawal behavior at one year of age is associated with delays in reaching language milestones in the EDEN mother-child cohort study. *Plos One, 11* (17).

Guedeney, A., Foucault, C., Bougen, E., Larroque,B.,& Mentré, F. (2008). Screening for risk factors of relational withdrawal behavior in infants aged 14–18 months. *European Psychiatry, 23*, 150–155.

Guedeney, A., Guedeney, N., Tereno, S., Dugravier, R., Greacen, T., Welniarz, B., Saias, T., Tubach, F., & the CAPDEP Study Group. (2011). Infant rhythms versus parental time: Promoting parent–infant synchrony. *Journal of Physiology-Paris, 105*, 195–200.

Guedeney, A., Le Foll, J. Vannier, L., Viaux-Savelon, S., & Wendland, J. (2014). *Petite enfance et psychopathologie*. Elsevier: Paris.

Guedeney, A., Marchand-Martin, L., Cote, S.J., Larroque, B., & the EDEN Mother–Child Cohort Study Group. (2012). Perinatal risk factors and social withdrawal behavior. *European Child & Adolescent Psychiatry, 21,* 185–191.

Guedeney, A., Pingault, J. B., Thorr, A., Larroque, B., The EDEN mother-child cohort study group (2014). Social withdrawal at 1 year is associated with emotional and behavioral problems at 3 and 5 years: the EDEN mother-child cohort study. *European Child and Adolescent Psychiatry*, *23* (12), 1181-1188.

Guedeney, A., Wendland, J., Dugravier, R., Saïas, T., Tubach, F., Welniarz, B., Guedeney, N., Greacen, T., Tereno, S., & Pasquet, B. (2013). Impact of a randomized home-visiting trial on infant social withdrawal in the CAPEDP prevention study. *Infant Mental Health Journal, 34* (6), 594-601.

Guedeney, N., & Fermanian, J. (1998). Validation study of the French version of the Edinburgh Postnatal Depression Scale (EPDS): New results about use and psychometric properties. *European Psychiatry, 13,* 83–89.

Habersaat, S., Monnier, M., Peter, C., Bolomey, L., Borghini, A., Despars, J., Pierrehumbert, B., Müller-Nix, C., Ansermet, F. & Hohlfeld, J. (2013). Early mother-child interaction and later quality of attachment in infants with orofacial cleft compared to infants without cleft. *The cleft Palate-Craniofacial Journal, 50* (6), 704-712.

Hall, P. (2012). Current considerations of the effects of untreated maternal perinatal depression and the national perinatal depression initiative. *Journal of Developmental Origins of Health and Disease, 3* (4), 293– 295.

Hanson, J. E., Lapidus, J., Zuniga, J., & Murphy, S. (2000). Childhood asthma in low-income Hispanic and non-Hispanic white families in a rural state: The Impact on Family Scale. *Children's Health Care*, *29*, 65-77.

Hair, J. F., Black, J. W., Babin, B. J., & Anderson, E. R. (2010). *Multivariate Data Analysis* (7th Ed.). Edinburgh: Pearson Education Limited.

Hardin-Jones, M. A., & Jones, D. L. (2005). Speech production patterns of preschoolers with cleft palate. *Cleft-Palate-Craniofacial Journal, 42*, 7-13.

Harper, D.C., & Richman, L.C. (1978). Personality profiles of physically impaired adolescents. *Journal of Clinical Psychology*, *34,* 636–42. Available in: https://www.ncbi.nlm.nih.gov/pubmed/151102

Hartley, C., Pretorius, K., Mohamed, A., Laughton, B., Madhi, S., Cotton, M.F. et al. (2010). Maternal postpartum depression and infant social withdrawal among human immunodeficiency virus (HIV) positive mother–infant dyads. Psychology, *Health & Medicine, 15* (3), 278–287.

Hendrick, S. S, & Hendrick, C. (1997). Love and satisfaction. In R. J. Sternberg & M. Hojjat (Eds.) *Satisfaction in close relationships*. (p. 56-78) New York: Guildford Press.

Hentges, F., Hill, J., Bishop, D.V.M., Goodacre, T., Moss, T., & Murray, L. (2011). The effect of cleft lip on cognitive development in school-aged children: a paradigm for examining sensitive period effects. *Journal of Child Psychology and Psychiatry, 52* (6), 704-712.

Hernández-Rief, M., Field, T., Diego, M., Vera, Y., & Pickens, J. (2005). Happy faces are habituated more slowly by infants of depressed mothers. *Infant Behavior and Development, 29* (1), 131-135.

Hoekstra-Weebers, J., Jaspers, J.P.C., Kamps, W.A., & Klip, E.C. (1998). Marital dissatisfaction, psychological distress, and the coping of parents of pediatric cancer patients. *Journal of Marriage and Family*, *60* (4), 1012–1021. doi: 10.2307/353642

Horwitz, A.V. (2007). Distinguishing distress from disorder as psychological outcomes of stressful social arrangements. *Health, 11,* 273-289.

Howard, L. (2005). Postnatal depression. *Clinical Evidence Concise, 13,* 410–412.

Hunfeld, J., Temples, A.m Passchier, J., Hazebroek, F., & Tibboel, D. (1999). Brief report: Parental burden and grief one year after the birth of a child with a congenital anomaly. *Journal of Pediatric Psychology, 24* (6), 515-520.

Hunt, O., Burden, D., Hepper, P., & Johnston, C. (2005). The psychosocial effects of cleft lip and palate: a systematic review. *European Journal of Orthodontics, 27* (3), 274–85. https://doi.org/10.1093/ejo/cji004

Hunt, O., Burden, D., Hepper, P., Stevenson, M., & Johnston, C. (2006). Self-reports of psychosocial functioning among children and young adults with cleft lip and palate. *Cleft Palate Craniofacial Journal*, *43*, 598–605.

Ilfeld, F.W. (1976). Further Validation of a Psychiatric Symptom Index in a Normal Population. *Psychological Report*, *39*, 1215-1228.

Ilfeld, F.W. (1978). Psychologic Status of Community Residents along Major Demographic Dimensions. *Archives of General Psychiatry, 35,* 716-724.

Institut National de la Statistique et des Études Économiques (INSEE). (2013). *Salaires dans le secteur privé et les entreprises publiques*. Available in: https://www.insee.fr/fr/statistiques/1370897

Izard, C.E., Huebner, R.R., Risser, D., McGinness, G., & Dougherty, L. (1980). The young infant's ability to produce discrete emotional expressions. *Developmental Psychology*, *16,* 132–140.

Jentink, J. Loane, M., Dolk, H., Barisic, I., Garne, E., Morris, J. K., & de Jong-van den Berg, L. (2010). Valproic acid monotherapy in pregnancy and major congenital malformations. *The New England Journal of Medicine, 362,* 2185–2193.

Johansson, B., & Ringsberg, K.C. (2004). Parents' experiences of having a child with cleft lip and palate. *Journal of Advanced Nursing*, *47*, 165–73.

Johnson, C. & Little, J. (2008). Folate intake, markers of folate status and oral clefts: is the evidence converging? *International Journal of Epidemiology, 37,* 1041–1058.

Jomeen, J. (2004). The importance of assessing psychological status during pregnancy, childbirth and the postnatal period as a multidimensional construct: a literature review. *Clinical Effectiveness in Nursing, 8,* 143-155.

Jones, M. C. (1988). Etiology of facial clefts: prospective evaluation of 428 patients. *Cleft Palate Journal, 25*, 16–20.

Jones, S., Statham, H., & Solomou, W. (2005). When expectant mothers know their baby has a fetal abnormality: exploring a crisis of motherhood through qualitative data-mining. *Journal of Social Work Research and Evaluation*, *62* (2), 195-206.

Josefsson, A., Berg, G., Nordin, C., & Sydsio, G. (2001). Prevalence of depressive symptoms in late pregnancy and postpartum. *Acta Obstetricia et Gynecologica Scandanavia, 80* (3), 251–255.

Josefsson, A., & Sydsjo. G. (2007). A follow-up study of postpartum depressed women: recurrent maternal depressive symptoms and

child behavior after four years. *Archives of Women's Mental Health*, *10*, 141-145.

Jugessur, A., & Murray, J.C. (2005). Orofacial clefting: recent insights into a complex trait. *Current Opinion in Genetics and Development*, *15* (3), 270–8.

Jugessur, A., Shi, M., Gjessing, H.K., Lie, R.T., Wilcox, A.J., Weinberg, C.R., Christensen, K., Boyles, A.L., Daack-Hirsch, S., Trung, T.N., Bille, C., Lidral, A.C., & Murray, J.C. (2009). Genetic determinants of facial clefting: analysis of 357 candidate genes using two national cleft studies from Scandinavia. *PLoS ONE*, *4* (4), e5385.

Kaasen, A., Helbig, A., Malt, U.F., Naes, T., Skari, H., & Haugen, G. (2010). Acute maternal social dysfunction, health perception and psychological distress after ultrasonographic detection of a fetal structural anomaly. *An International Journal of Obstetrics and Gynecology*, *117,* 1127-1138.

Kapp-Simon, K.A., Simon, D.J., & Kristovich, S. (1992). Self-perception, social skills, adjustment, and inhibition in young adolescents with craniofacial anomalies. *Cleft Palate Craniofacial Journal, 29*, 352–356.

Kapp-Simon, K.A., & McGuire, D.E. (1997). Observed social interaction patterns in adolescents with and without craniofacial conditions. *Cleft Palate Craniofacial Journal*, *34* (5), 380–4.

Kasten, E.F., Schmidt, S.P., Zickler, C.F., Berner, E., Damian, L.A., Christian, G.M., Workman, H., Freeman, M., Farley, M.D., & Hicks, T.L. (2008). Team care of the patient with cleft lip and palate. *Current Problems in Pediatric and Adolescent Health Care*, *38,* 138–158.

Katz-Wise, S., Priess, H., & Hyde, J. (2010). Gender-role attitudes and behavior across the transition to parenthood. *Developmental Psychology, 46,* 18–28. doi: 10.1037/a0017820.

Kline, R. B. (2011). *Principles and Practice of Structural Equation Modeling* (5th Ed.). New York: The Guilford Press.

Kolk, A.M., Schipper, J.L., Hanewald, G.J., Casari, E.F., & Fantino, A.G. (2000). The Impact-on-Family Scale: a test of invariance across culture. *J Pediatr Psychol, 2*5(5), 323-9.

Koomen, H.M. & Hoeksma, J.B. (1992). Maternal interactive behaviour towards children with and children without cleft lip and palate. *Early Development and Parenting Journal*, *1,* 169–181.

Koomen, H.M., & Hoeksma, J.B. (1993). Early hospitalization and disturbances of infant behavior and the mother–infant relationship. *Journal of Child Psychology and Psychiatry, 34*, 917–34.

Kramer, F. J., Beathge, C., Sinikovic, B., & Schliephake, H. (2007). An analysis of quality of life in 130 families having a small children with cleft lip/palate using the impact on family scale. International *Journal of Oral and Maxillofacial Surgery, 36* (12), 1146-1152.

Kummer, A.W. (2014). *Cleft palate and craniofacial anomalies: Effects on speech and resonance*. (3rd Ed.). New York: Delmar Cengage Learning.

Kuttenberger, J., Ohmer, J.N., & Polska, E. (2010). Initial counselling for cleft lip and palate: parents' evaluation, needs and expectations. *International Journal of Oral and Maxillofacial Surgery*, *39,* 214–20.

Lalor, J., Begley, C., & Galavan, E. (2009). Recasting hope: A process of adaptation following fetal anomaly. *Social Science and Medicine, 68,* 462-472.

Langlois, J.H., Ritter, J.M., Casey, R.J., & Sawin, D.B. (1995). Infant attractiveness predicts maternal behaviors and attitudes. *Developmental Psychology*, *31*, 464–472.

Lansdown, R. (1981). Cleft lip and palate: a prediction of psychological disfigurement? *British Journal of Orthodontics*, *8* (2), 83–88.

Lansdown, R., Lloyd, J., & Hunter, J. (1991). Facial deformity in childhood: severity and psychological adjustment. *Child Care, Health and Development*, *17*, 165–71.

Lawoko, S., & Soares, J. (2006). Psychosocial morbidity among parents of children with congenital heart disease: a prospective longitudinal study. *Heart and Lung: The Journal of Acute and Critical Care*, *35* (5), 301-314

Leemreis, W. H., Okkerse, J. M., de Laat, P., Madern, G., van Adrichem, L., Verhulst, F., & Oranje, A. (2014). Educational paper: Parenting a child with a disfiguring condition –how (well) do parents adapt? European Journal of Pediatrics, 173, 699-709.

Letourneau, N. L., Dennis, C.-L., Benzies, K., Duffett-Leger, L., Stewart, M., Tryphonopoulos, P. D., Este, D., & Watson, W. (2012). Postpartum depression is a family affair: Addressing the impact on mothers, fathers, and children. *Issues in Mental Health Nursing*, 33, 445–457. doi:10.3109/01612840.2012.673054

Lewis, J. (1998a). The transition to parenthood: I. The rating of prenatal marital competence. *Family Process*, *27*, 149-165.

Lewis, J. (1998a). The transition to parenthood: II. Stability and change in marital structure. *Family Process*, *27,* 273-283.

Lieberman, A.F., & Zeanah, C.F. (1995). Disorders of attachment in infancy. *Child and Adolescent Psychiatric Clinics of North America, 4,* 571–587.

Little, J., Cardy, A. & Munger, R. G. (2002). Tobacco smoking and oral clefts: a meta-analysis. *Bulletin of the World Health Organization, 82* (3), 213–218.

Locock, L., & Alexander, J. (2006). Just a bystander? Men's place in the process of fetal screening and diagnosis. *Social Sciences and Medicine, 62*, 1349-1359

López-Camelo, J. S., Castilla E. E. & Orioli, I. M. (2010). Folic acid flour fortification: impact on the frequencies of 52 congenital anomaly types in three South American countries. *American Journal of Human Genetics, 152A*, 2444–2458.

Maarse, W., Berge, S.J., Pistorius, L., van Barneveld, T., Kon, M., Breugem, C., & Mink van der Molen, A. B. (2010). Diagnostic accuracy of transabdominal ultrasound in detecting prenatal cleft lip and palate: a systematic review. *Ultrasound in Obstetrics and Gynecology*, *35*, 495–502.

Madsen, S.A., & Juhl, T. (2007). Paternal depression in the postnatal period assessed with traditional and male depression scales. *International Journal of Men's Health and Gender, 4*, 26–31.

Maes, S., Demey, A., & Appelboom-Fondu, J. (1998). Impact of ultrasound for facial cleft on mother-child relationships. *Annals of the New York Academy of Sciences, 847*, 249–51.

Mäntymaa, M. (2006). *Early mother–infant interaction: Determinants and predictivity.* Unpublished dissertation. University of Tampere. Available in: http://acta.uta.fi

Mäntymaa, M., Puura, K., Luoma, I., Kaukonen, P., Salmelin, R.K., & Tamminen, T. (2008). Infants' social withdrawal and parents' mental health. *Infant Behavior & Development, 31* (4), 606–613.

Marazita, M. L., Field, L., Cooper, M., Tobias, R., Maher, B., Peanchitlertkajorn, S., & Liu, Y. (2002). Nonsyndromic cleft lip with or without cleft palate in China: assessment of candidate regions. *Cleft Palate Craniofacial Journal, 39*, 149–156.

Maris, C.L., Endriga, M.C., Speltz, M.L., Jones, K., & DeKlyen, M. (2000). Are infants with orofacial clefts at risk for insecure mother–child attachments? *Cleft Palate-Craniofacial Journal*, *37,* 257–265.

Martelli, D., Coletta, R., Oliveira, E.A., Swerts, M.S.O., Rodrigues, L.A.M., Oliveira, M.C., & Martelli, H. (2015). Association between maternal smoking, gender, and cleft lip and palate. *Brazilian Journal of Otorhinolaryngology, 81 (5),* 514-519.

Matijasevich, A., Murray, J., Cooper, P. J., Anselmi, L., Barros, A. J. D., Barros, F. C., & Santos, I. S. (2015). Trajectories of maternal depression and offspring psychopathology at 6 years: 2004 Pelotas cohort study. *Journal of Affective Disorders, 174*, 424–431. doi:10.1016/j.jad.2014.12.012

Matthey, S., Barnett, B., Kavanagh, D. J., & Howie, P. (2001). Validation of the Edinburgh Postnatal Depression Scale for men, and comparison of item endorsement with their partners. *Journal of Affective Disorders, 64,* 175-184.

Matthey, S., Guedeney, A., Starakis, N., & Barnett, B. (2005). Assessing the social behavior of infants: Use of the ADBB scale and relationship to mother's mood. *Infant Mental Health Journal, 26,* 442–458.

Matthey, S., Crncec, R., Hales, A., & Guedeney, A. (2013). A description of the Modified Alarm Distress Baby Scale (m-ADBB): An instrument to assess for infant social withdrawal. *Infant Mental Health Journal, 34* (2), 602-609.

Mazer, P., Gischler, S., Koot, H., Tibboel, D., van Dijk, M., & Duivenvoorden, H. (2008). Impact of a child congenital anomalies on parents (ICCAP) questionnaire; a psychometric analysis. *Health and Quality of Life Outcomes*, *6*:102.

McCubbin, H. I., Thompson, A. I., & McCubbin, M. A. (1996). Resiliency in families: A conceptual model of family adjustment and adaptation in response to stress and crisis. In H. I. McCubbin, A. I. Thompson, & M. A. McCubbin (Eds.), *Familiy assessment: Reciliency, coping and adaptation –inventories for research and practice*- (p. 1–64). Madison, WI: University of Wisconsin Publishers.

McGrattan, K. & Ellis, C. (2013). Team-Oriented care for Orofacial clefts: a review of the literature. *The Cleft Palate-Craniofacial Journal, 50* (1), 13-18.

McHale, J. (2007). *Charting the bumpy road of coparenthood: Understanding the challenges of family life*. Washington, DC: Zero To Three.

McHale, J., Lauretti, A., Talbot, J., & Pouquette, C. (2002). Retrospect and prospect in the psychological study of coparenting and family group process. In J. McHale & W. Grolnick (Eds.), *Retrospect and prospect in the psychological study of families* (p. 127-165). Manwah, N. J.: Erlbaum.

McWilliams, B. J. (1982). Social and psychological problems associated with cleft palate. *Clinics in Plastic Surgery, 61*, 564-569.

Meltzoff, A. N., & Moore, M. K. (1977). Explaining facial imitation: A theoretical model. *Early Development and Parenting*, *6,* 179-192.

Messias, D., Gilliss, C., Sparacino, P., Tong, E., & Foote, D. (1995). Stories of transition: Parents recall the diagnosis of congenital heart defect. *Family Systems Medicine, 13*, 367–377.

Milne, L., Greenway, P., Guedeney, A., & Larroque, B. (2009). Long term developmental impact of social withdrawal in infants. *Infant Behavior & Development, 32*, 159–166.

Minde, K., Perrotta, M., & Marton, P. (1985). Maternal caretaking and play with full-term and premature infants. *Journal of Child Psychology and Psychiatry, 26,* 231–244.

Mirowsky, J., & Ross, C.E. (2002). Selecting outcomes for the sociology of mental health: Issues of measurement and dimensionality. *Journal of Health and Social Behavior*, *43*, 152-170.

Mitchell, L.E. (2002). Mode of inheritance of oral clefts. In: D.F. Wyszyski (Ed.), *Cleft Lip and Palate: From Origin to Treatment* (p. 234–239). Oxford University Press.

Moe, V., Braarud, H., Wentzel-Larsen, T., Slinning, K., Vannebo, U. T., Guedeney, A., Heimann, M., Rostad, A. M., & Smith, L. (2016). Precursor of social emotional functioning among full-term and preterm infants at 12 months: Early infant withdrawal behavior and symptoms of maternal depression. *Infant Behavior and Development, 44,* 159-168.

Molteno, C., Jacobson, J. L., Carter C., Dodge, N., & Jacobson, S. (2014). Infant emotional withdrawal: A precursor of affective and cognitive disturbance in fetal alcohol spectrum disorders. *Alcoholism: Clinical and Experimental Research, 38* (2), 479-488.

Montirosso, R., Borgatti, R., Trojan, S., Zanini, R., & Tronick, E. (2010). A comparison of dyadic interactions and coping with still-face in healthy pre-term and full-term infants. *British Journal of Developmental Psychology, 28,* 347-368.

Montirosso, R., Fedeli, C., Murray, L., Morandi, F., Brusati, R., Perego, G., & Borgatti, R. (2012). The role of negative maternal affective states and infant temperament in early interactions between infants with cleft lip and their mothers. *Journal of Pediatric Psychology, 37* (2), 241-250.

Mossey, P., Davies, J. A. & Little, J. (2007). Prevention of orofacial clefts: does pregnancy planning have a role? *The Cleft Palate-Craniofacial Journal, 434,* 244–250.

Mossey P. & Little, J. (2009). Addressing the challenges of cleft lip and palate research in India. *Indian Journal of Plastic Surgery, 42*, 9–18.

Mossey, P., Little, J., Munger, R. G., Dixon, M. J. & Shaw, W. C. (2009). Cleft lip and palate. *Lancet, 374*, 1773–1785.

Munger, R. G., Tamura, T., Johnston, K. E., Feldkamp, M., Pfister, R., Botto, L., & Carey, J. (2009). Plasma zinc concentrations of mothers and the risk of oral clefts in their children in Utah. *Clinical and Molecular Teratology, 85*, 151–155.

Munro, I. R. (1995). A description of craniofacial anomalies: The mechanism and rationale of surgery. In R. Eder (Ed.), *Cranio-facial anomalies, psychological perspectives*. New York: Springer-Verlag.

Murphy, N. A., Christian, B., Caplin, D. A., & Young, P. C. (2007). The health of caregivers for children with disabilities: Caregiver perspectives. *Child: Care Health Development*, *33*, 180–187.

Murray, J. C. (2002). Gene/environment causes of cleft lip and/ or palate. *Clinical Genetics, 61,* 248–256.

Murray, L. (1992). The impact of postnatal depression on infant development. *Journal of Child Psychology and Psychiatry*, *33*, 534-561.

Murray, L., Arteche, A., Bingley, C., Hentges, F., Bishop, D.V.M., Dalton, L., Goodacre, T. & Hill., J. (2010). The effect of cleft lip on socio-emotional functioning in school-aged children. *Journal of Child Psychology and Psychiatry, 51*, 94-103.

Murray, L., Arteche, A., Fearon, P., Halligan, S., Goodyer, I., & Cooper, P. (2011). Maternal postnatal depression and the development of depression in offspring up to 16 years of age. *Journal of the American Academy of Child & Adolescent Psychiatry, 50*, 460–470. doi:10.1016/j.jaac.2011.02.001

Murray, L. & Carothers, A. D. (1990). Validation of the Edinburgh Postnatal Depression Scale on a community sample. *British Journal of Psychiatry, 166,* 595-600.

Murray, L., & Cooper, P. (2003). Intergenerational transmission of affective and cognitive processes associated with depression: infancy and the preschool years. In: I. Goodyer (Ed.), *Unipolar depression: a lifespan perspective.* Oxford: Oxford University Press.

Murray, L., Fiori-Cowley, A., Hooper, R., & Cooper, P. (1996). The impact of postnatal depression and associated adversity on early mother– infant interactions and later infant outcome. *Child Development, 67*, 2512–2526.

Murray, L., Hentges, F., Hill, J., Karpf, J., Mistry, B., Kreutz, M., Woodall, P., Moss, T., & Goodacre, T. (2008). The effect of cleft lip and palate, and the timing of lip repair on mother-infant interactions and infant development. *Journal of Child Psychology and Psychiatry*, *49,* 115–123.

Murray, L., & Trevarthen, C. (1985). Emotional regulation of interactions between 2 month-olds and their mothers. In T. M. Field & N. A. Fox (Eds.), *Social perception in infants* (p. 177-197). Norwood, N.J.: Ablex.

Musters, C., McDonald, E., & Jones, I. (2008). Management of postnatal depression. *British Medical Journal, 337*, a736.

Nackashi, J. A., Dedlow, R.E., & Wood-Dixon, V. (2002). Health care for children with cleft lip and palate: comprehensive services, and infant feeding. In: Wyszynski, D.F. (Ed.). *Cleft Lip and Palate from Origin to Treatment* (p. 127-158). New York, NY: Oxford University Press.

Neiman, G. S. & Savage, H. E. (1997). Development of infants and toddlers with clefts from birth to three years. *Cleft Palate Craniofacial Journal, 34,* 218-225.

Nelson, P., Glenny, A.M., Kirk, S., & Caress, A.L. (2012). Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child: Care, Health and Development*, *38,* 6–20.

Nelson, P., O'Leary, C., & Weinman, J. (2009). Causal attributions in parents of babies with a cleft lip and/or palate and their association with psychological well-being. *Cleft Palate Craniofacial Journal, 46*, 425–434.

Nes, R.B., Røysamb, E., Hauge, L.J., Kornstad, T., Landolt, M.A., Irgens, L.M., Eskedal, L., Kristensen, P., & Vollrath, M.E. (2014). Adaptation to the birth of a child with a congenital anomaly: a prospective longitudinal study of maternal well-being and psychological distress. Developmental Psychology, 50 (6), 1827-1839.

Nicely, P., Tamis-LeMonda, C.S., & Grolnick, W.S. (1999). Maternal responsiveness to infant affect: Stability and prediction. *Infant Behavior and Development, 22,* 103–117.

Nidey, N., Moreno, L. M., Marazita, M., & Wehby, G. L. (2015). Psychosocial well-being of parents of children with oral clefts. *Child: Care, Health and Development*, *42*, 42-50.

Nomaguchi, K., & Milkie, M. (2003). Costs and rewards of children: the effects of becoming a parent on adults' lives. *Journal of Marriage and Family, 65* (2), 356-374.

Nunnally, J.C. (1967). *Psychometric theory*. New York: McGraw Hill.

Nusbaum, R., Grubs, R.E., Losee, J.E., Weidman, C., Ford, M.D., & Marazita, M.L. (2008). A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period. *Journal of Genetic Counseling, 17,* 336–350.

Oliver, M., Yulitta, H., & Guedeney, A. (2016). Análisis psicométrico de la Escala de alarma sobre el retraimiento del bebé de uso pediátrico*. Acta Psiquiátrica y Psicológica de América Latina, 62* (2), 77-84.

Panksepp, J. (2006). Emotional endophenotypes in evolutionary psychiatry. *Progress in Neuro-Psychopharmacology & Biological Psychiatry*, *30,* 774–784.

Papp, L.M., Goeke-Morey, M.C., & Cummings, E.M. (2007). Links between spouses' psychological distress and marital conflict in the home. *Journal of Family Psychology*, *21,* 533–537.

Patrick, D.L., Topolski, T.D., Edwards, T.C., Aspinall, C.L., Kapp-Simon, K.A., Rumsey, N.J., Strauss, R.P., & Thomas, C.R. (2007). Measuring the quality of life of youth with facial differences. *Cleft Palate Craniofacial Journal, 44,* 538–547.

Paulson, J. F., & Bazemore, S. D. (2010). Prenatal and postpartum depression in fathers and its association with maternal depression. A meta-analysis. *Journal of the American Medical Association, 303* (19), 1961-1969. doi:10.1001/jama.2010.605

Pelchat, D., Bisson, J., Bois, C., & Saucier, J. (2003). The effects of early relational antecedents and other factors on the parental sensitivity of mothers and fathers. *Infant and Child Development*, *12*, 27–51.

Pelchat, D., Lefebvre, H., Proulx, M., & Reidy, M. (2004). Parental satisfaction with an early intervention program. *Journal of Perinatal and Neonatal Nursing, 18* (2), 128–144.

Pelchat, D., Ricard, N., Bouchard, J.M., Perreault, M., Saucier, J.F., Berthiaume, M., & Bisson, J. (1999). Adaptation of parents in relation to their 6-month-old infant's type of disability. *Child: Care, Health and Development, 25,* 377-397.

Pérez-Martínez, C. & Guedeney, A. (2015). Retraimiento social en la primera infancia. Implicaciones en el desarrollo del psiquismo. *Revista Uruguaya de Psicoanálisis, 120*, 120-132.

Peterson-Falzone, S.J., Hardin-Jones, M.A., & Karnell, M.P. (2010). *Cleft palate speech*. (4th Ed.), Iowa City, Iowa: Mosby. Available in https://www.elsevier.com/books/cleft-palate-peech/peterson-falzone/978-0-323-04882-8

Pope, A. W., Tillman, K., & Snyder, H. T. (2005). Parenting stress in infancy and psychosocial adjustment in toddlerhood: A longitudinal study of children with craniofacial anomalies. *The Cleft Palate-Craniofacial Journal, 42,* 556–559.

Pope, A.W., & Ward, J. (1997). Factors associated with peer social competence in preadolescents with craniofacial anomalies. *Journal of Pediatric Psychology, 22,* 455–469.

Porter, F. (2006). Cholesterol precursors and facial clefting. *J. Clin. Invest. 116*, 2322–2325.

Powell, G.F., & Low, J. (1983). Behavior in nonorganic failure to thrive. *Developmental and Behavioral Pediatrics*, *4*, 26–33.

Préville M., Boyer R., Potvin L., Perrault C., & Légaré G. (1992). *La détresse psychologique: détermination de la fiabilité et de la validité de la mesure utilisée dans l'enquête Santé Québec,* Enquête Santé Québec 87. Les cahiers de recherche nº7. Montreal: Ministère de la Santé et des Services Sociaux.

Puura, K., Guedeney, A., Mäntymaa, M., & Tamminnen, T. (2007). Detecting infants in need: Are complicated measures really necessary? *Infant Mental Health Journal*, *28* (4), 409–421.

Puura, K., Mäntymaa, M., Luoma, I., Kaukonen, P., Guedeney, A., Salmelin, R., & Tamminen, T. (2010). Infant's social withdrawal symptoms assessed with a direct method in primary health care. *Infant Behavior & Development*, *33*, 579–588.

Puura, K., Mäntymaa, M., Leppänen, J., Peltola, M., Salmellin, R., Luoma, I., Latva, R., & Tamminen, T. (2013). Associations between maternal interaction behavior, maternal perception of infant temperament, and infant social withdrawal. *Infant Mental Health Journal*, *34* (6), 586-593.

Rahman, A., Iqbal, Z., Bunn, J., Lovel, H., & Harrington, R. (2004). Impact of maternal depression on infant nutritional status and

illness: a cohort study. *Archives of General Psychiatry*, *61,* 946–952.

Ramchandani, P., Stein, al., Evans, J., O'Connor, & the ALSPAC study team (2005). Paternal depression in the postnatal period and child development: a prospective population study. *The Lancet, 365,* 2201-2205.

Ramchandani, P., Psychogiou, L., Vlachos, H., Iles, J., Sethna, V., Netsi, E., & Lodder, A. (2011). Paternal depression: An examination of its links with father, child and family functioning in the postnatal period. *Depression and Anxiety*, *28,* 471-477.

Ramstad, T., Ottem, E., & Shaw, W.C. (1995). Psychosocial adjustment in Norwegian adults who had undergone standardized treatment of complete cleft lip and palate. I. Education, employment and marriage. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, *29,* 251–257.

Ray, J.G., Vermeulen, M.J., Wyatt, P.R. & Cole, D.E. (2003). Association between folic acid food fortification and congenital orofacial clefts. *Journal of Pediatrics, 143,* 805–807.

Rey-Bellet, C. & Hohlfeld, J. (2004). Prenatal diagnosis of facial clefts: evaluation of a specialized counselling. *Swiss Medical Weekly*, *134,* 640-644.

Richman, L.C., & Ryan, S.M. (2003). Do the reading disabilities of children with cleft fit into current models of developmental dyslexia? *Cleft Palate Craniofacial Journal, 40,* 154–157.

Richman, L.C., Wilgenbusch, T., & Hall, T. (2005). Spontaneous verbal labeling: visual memory and reading ability in children with cleft. *Cleft Palate Craniofacial Journal, 4,* 565–569.

Ridner, S.H. (2004). Psychological distress: concept analysis. *Journal of Advanced Nursing, 45,* 536-545.

Roberts, R., Mathias, J. L., & Wheaton, P. (2012). Cognitive functioning in children and adults with nonsyndromal cleft lip and/or palate: A meta-analysis. *Journal of Pediatric Psychology*, *37* (7), 786–797.

Robertson, J., & Bowlby, J. (1952). Responses to young children to separation from their mothers. *Courrier du Centre International de l'Enfance, 2,* 131–142.

Robertson, E., Celasun, N., and Stewart, D.E. (2003). Risk factors for postpartum depression. Postpartum depression: Literature review of risk factors and interventions. In D.E. Stewart, E. Robertson, C.L. Dennis, S.L. Grace, & T. Wallington, *Maternal mental health and child health and development* (p. 13-70). Toronto: University Health Network Women's Health Program.

Rochette, J., & Mellier, D. (2007). Transformation des souffrances de la dyade mère-bébé dans la première année postpartum: stratégies préventives pour un travail en réseau, *Devenir, 19* (2), 81-108.

Rosenn, D.W., Loeb, L.S., & Bates, M. (1980). Differentiation of organic from nonorganic failure to thrive syndrome in infancy. *Pediatrics*, *66*, 698–704.

Rubin, K.H., Coplan, R., & Bowker, J.C. (2009). Social withdrawal in childhood. *Annual Review of Psychology*, *60*, 141–171.

Rumsey, N., & Harcourt, D. (2005). *The Psychology of Appearance.* Berkshire: Open University Press.

Russell, A., Mize, J., & Bissaker, K. (2002). Parent–child relationships. In P.K. Smith & C.H. Hart (Eds.), H*andbook of childhood social development* (p. 205–222). Oxford, England: Blackwell.

Rvachew, S., Slawinski, E. B., Williams, M., & Green, C. (1999). The impact of early onset of otitis media on babbling and early language development. *Journal of the Acoustical Society of America, 105*, 467-475.

Sameroff, A.J. (1993). Models of development and developmental risk. In C.H. Zeanah, Jr. (Ed.), *Handbook of infant mental health*. New York: Guilford Press.

Sameroff, A.J., & Emde, R.N. (1989). Relationship disturbances in context. In A.J. Sameroff & R.N. Emde (Eds.), *Relationship disturbances in early childhood* (p. 221–238). New York: Basic Books.

Sameroff, A.J., & Fiese, B.H. (1990). Transactional regulation and early intervention. In S.J. Meisels & J.P. Shonkoff (Eds.), *Handbook of early childhood intervention*. New York: Cambridge University Press.

Sameroff, A.J., & Fiese, B.H. (2000). Models of development and developmental risk. (p. 3-19). In C. H. Zeanah (Ed.). *Handbook of infant mental health*. New York: The Guilford Press.

Sank, J.R., Berk, N.W., Cooper, M.E., & Marazita, M.L. (2003) Perceived social support of mothers of children with clefts. *Cleft Palate Craniofacial Journal, 40* (2), 165–171

Sausse, S. (1996). *Le miroir brisé*. Paris: Calmann-Lévy.

Scherer, N. J., D' Antonio, L. L., & Kalbfleisch, J. H. (1999). Early speech and language development in children with velocardiofacial syndrome. *American Journal of Medical Genetics*, *88,* 714-723.

Schlenker, M., Maury, M., Montoya, P., & Visier, J.P. (1998a). L'annonce du diagnostic de malformation faciale: importance d'un travail de soutien psychologique auprès des familles et des équipes soignantes. *Psychiatrie de l'enfant*, *41,* 37–86.

Schlenker, M., Montoya, P., Maury, M., & Visier, J.P. (1998b). Le diagnostic de malformation faciale: intérêts de l'annonce anténatale. In: P. Mazet, & S. Lebovici (Eds.), *Psychiatrie périnatale. Parents et bébés: du projet d'enfant aux premiers mois de vie* (p.169–185). PUF: Paris.

Schore, A. (2001). Effects of a secure attachment relationship on right brain development, affect regulation, and infant mental health. *Infant Mental Health Journal*, *22*, 7–66.

Schulz, M.S., Cowan, C.P., & Cowan, P.A. (2006). Promoting healthy beginnings: a randomized controlled trial of a preventive intervention to preserve marital quality during the transition to parenthood. *Journal of Consulting and Clinical Psychology*, *74*, 20-31.

Shahrukh Hashmi, S., Gallaway, M. S., Waller, D. K., Langlois, P. H. & Hecht, J. T. (2010). National Birth Defects Prevention Study. Maternal fever during early pregnancy and the risk of oral clefts. *Birth Defects Research. Part A Clinical and Molecular Teratology, 288,* 186–194.

Sharp, M. C., Strauss, R. P., & Lorch, S. C. (1992). Communicating medical bad news: Parent's experiences and preferences. *Journal of Pediatrics, 4* (121), 539-546.

Shaw, W.C., Semb, G., Nelson, P., Brattström, V., Mølsted, K., & Prahl-Andersen, B. (2001). The Eurocleft Project 1996-2000: Overview. *Journal of Cranio-Maxillo-Facial Surgery*, *29*, 131-142.

Shi, M., Christensen, K., Weinberg, C., Romitti, P., Bathum, L., Lozada, A., Morris, R.W., Lovett, M., & Murray, J. (2007). Orofacial cleft risk is increased with maternal smoking and specific detoxification-gene variants. *American Journal of Human Genetics, 80,* 76–90.

Shi, M., Wehby, G. L. & Murray, J. C. (2008). Review on genetic variants and maternal smoking in the etiology of oral clefts and other birth defects. *Birth Defects Research. Part C Embryo Today, 84,* 16–29.

Shields, C.G., & Rousseau, S.J. (2004). A pilot study of an intervention for breast cancer survivors and their spouses. *Family Process, 43,* 95—107.

Skari, H., Malt, U., Bjornland, K., Egeland, T., Haugen, g., Skreden, M., Dalholt, B., Bjornland, O., & Emblem, R. (2006). Prenatal diagnosis of congenital malformations and parental psychological distress: a prospective longitudinal cohort study. *Prenatal Diagnosis, 26*, 1001-1009.

Slade, P., Emerson, D.J.M., & Freedlander, E. (1999). A longitudinal comparison of the psychological impact on mothers of neonatal and 3 month repair of cleft lip. *British Journal of Plastic Surgery, 52*, 1–5.

Slutsky, H. (1969). Maternal reaction and adjustment to birth and care of cleft palate child. *Cleft Palate Journal*, *6*, 425–429

Snow, C.W. (1998). *Infant development* (2nd Ed.). Upper Saddle River, NJ: Prentice Hall.

Solberg, O., Dale, M.T., Holmstrom, H., Eskedal, L.T., Landolt, M.A., & Vollrath, M.E: (2011). Long-term symptoms of depression and anxiety in mothers of infants with congenital heart defects. *Journal of Pediatric Psychology*, *36,* 179–187. doi: 10.1093/jpepsy/jsq054

Solomon, J., George, C., 1999. The measurement of attachment security in infancy and childhood. In: Cassidy, J., Schavers, J. (Eds.), *Handbook of Attachment* (p. 287-318). New York, N.Y.: Guildford Press.

Spanier, G. B. (1976). Measuring dyadic adjustment: New scales for assessing the quality of marriage and similar dyads. *Journal of Marriage and the Family*, *38*, 15-30.

Speltz, M. L., Armsden, G. C., & Clarren, S. S. (1990). Effects of craniofacial birth defects on maternal functioning postinfancy. *Journal of Pediatric Psychology*, *15* (2), 177–196. http://dx.doi.org/10.1093/jpepsy/15.2.177

Speltz, M., Endriga, M., Fisher, P., & Mason, C. (1997). Early predictors of attachment in infants with cleft lip and/or palate. *Child Development, 68,* 12–25.

Speltz, M., Endriga, M., Hill, S., Maris, C., Jones, K., & Omnell, M. (2000). Brief report: Cognitive and psychomotor development of infants with oral-facial clefts. *Journal of Paediatric Psychology, 25,* 185–190.

Speltz ML, Goodell EW, Endriga MC, & Clarren SK. (1994). Feeding interactions of infants with unrepaired cleft lip and/or palate. *Infant Behavior Development*, *17*, 131–139.

Speltz, M.L., Morton, K., Goodell, E.W., & Clarren, S.K. (1993). Psychological functioning of children with craniofacial anomalies and their mothers: follow-up from late infancy to school entry. *Cleft Palate Craniofacial Journal*, *30*, 482-489.

Spitz, R.A. (1946). Anaclitic depression. *Psychoanalytical Study of the Child, 2*, 313–341.

Stein, R., & Reissman, C. (1980). The Development of an Impact-on-Family Scale: Preliminary findings. *Medical Care*, *18* (4), 465-472.

Stern, D.N. (1977). *The first relationship: Infant and mother*. Cambridge, MA: Harvard University Press.

Stern, D.N. (1985). *The interpersonal world of the infant. A view from psychoanalysis and developmental psychology*. New York: Basic Books.

Stock, N. M., & Feragen, K. B. (2016) Psychological adjustment to cleft lip and/or palate: A narrative review of the literature. *Psychology and Health,* 31 (7), 777-813. Available in: http://eprints.uwe.ac.uk/27945

Stock, N. M., & Rumsey, N. (2015). Parenting a child with a cleft: The father's perspective. *The Cleft Palate-Craniofacial Journal, 52*, 31–43. http://dx.doi.org/10.1597/13-035.

Strauss, R.P. (2002). Beyond easy answers: prenatal diagnosis and counseling during pregnancy. *The Cleft Palate-Craniofacial Journal, 39*, 164–168.

Strauss, R.P., Broder, H., & Helms, R.W. (1988). Perception of appearance and speech by adolescent patients with cleft lip and palate and their parents. *Cleft Palate Journal, 25,* 335–42.

Tabachnick, B. G., & Fidell, L. S. (2013). *Using multivariate statistics* (6th Ed.). New Jersey: Pearson Education Inc.

Tauber, M., Boulanouar, K., Diene, G., Cabal-Berthoumieu, S., Ehlinger, V., et al. (2017). The Use of Oxytocin to Improve Feeding and Social Skills in Infants with Prader–Willi Syndrome. *Pediatrics, 139* (2):e20162976

Teti, D.M., Gelfand, D.M., Messinger, D.S., & Isabella, R. (1995).Maternal depression and the quality of early attachment: An examination of infants, preschoolers, and their mothers. *Developmental Psychology, 31* (3), 364–376.

Topolski, T.D., Edwards, T.C., & Patrick, D.L. (2005). Quality of life: how do adolescents with facial differences compare with other adolescents? *The Cleft Palate-Craniofacial Journal, 42*, 25–32.

Thomas, P.T., Turner, S.R., Rumsey, N., Dowell, T., Sandy, J.R. (1997). Satisfaction with facial appearance among subjects affected by a cleft. *The Cleft Palate-Craniofacial Journal, 34*, 226–231.

Tizard, B. (1977). *Adoption: A second chance*. New York: Free Press.

Tizard, B., & Rees, J. (1975). The effects of early institutional rearing on the behavior problems and affectional relationships of four-year-old children. *Journal of Child Psychology and Psychiatry, 16,* 61–73.

Trevarthen, C., & Aitken, K.J. (2001). Infant intersubjectivity: Research, theory, and clinical applications. *Journal of Child Psychology and Psychiatry, 42*, 3–48.

Tronick, E.Z. (1989). Emotions and emotional communication in infants. *American Psychologist*, *44*, 112–126.

Tronick, E.Z., Als, H., & Brazelton,T.B. (1980). Monadic phases: A structural description analysis of infant-mother face-to-face interaction. *Merrill-Palmer Quarterly*, *26*, 3-24.

Tronick, E.Z., & Gianino, A.F. (1986). The transmission of maternal disturbance to the infant. In E.Z. Tronick & T. Field (Eds.), *Maternal depression and infant disturbance* (p. 5–11). *New Directions for Child Development*. San Francisco: Jossey-Bass.

Tronick, E.Z., & Reck, C. (2009). Infants of depressed mothers. *Harvard Review of Psychiatry*, 17 (2), 147-156.

Tronick, E.Z., & Weinberg, M. K. (1997). Depressed mothers and infants: Failure to form dyadic states of consciousness. In L. Murray & P. J. Cooper (Eds.), *Postpartum depression and child development* (p. 54-81). New York: Guilford Press.

Turner, S.R., Rumsey, N., & Sandy, J.R. (1998). Psychological aspects of cleft lip and palate. *European Journal of Orthodontics*, *20*, 407-415.

Turner, S.R., Thomas, P.W., Dowell, T., Rumsey, N., & Sandy, J.R. (1997). Psychological outcomes amongst cleft patients and their families. *British Journal of Plastic Surgery, 50,* 1–9

Van der Waerden, J., Galéra, C., Larroque, B., Saurel-Cubizolles, M.-J., Sutter-Dallay, A.-L., & Melchior, M. (2015). Maternal depression trajectories and children's behavior at age 5 years. *Journal of Pediatrics*. Advance online publication. doi:10.1016/j.jpeds.2015.03.002

Van Egeren, L. (2003). Prebirth predictors of coparenting experiences in early infancy. *Infant Mental Health Journal*, *24* (3), 278-295.

Van Schoors, M., Caes, L., Alderfer, M., Goubert L., & Verhofstadt, L. (2016). Couple functioning after pediatric cancer diagnosis: a systematic review. *Psycho-Oncology*, *26*, 608-616.

Van Staden, F., & Gerhardt, C. (1995). Mothers of children with facial cleft deformities: reactions and effects. *South African Journal of Psychology, 25*, 39–46.

Viaux-Savelon, S., Rosenblum, O., Mazet, P., Dommergues, M., & Cohen, D. (2007). La surveillance échographique prénatale des grossesses à suspicion de malformation: étude du retentissement sur les représentations maternelles. *Neuropsychiatrie de l'Enfance*

et de l'Adolescence, 55, 413–423. https://doi.org/10.1016/j.neurenf.2007.07.022

Vidair, H.B., Reyes, J.A., Shen, S., Parrilla-Escobar, M.A., Heleniak, C.M., Hollin, I.I. et al. (2011). Screening parents during child evaluations: Exploring parent and child psychopathology in the same clinic. *Journal of the American Academy of Child & Adolescent Psychiatry*, *50* (5), 441–450.

Wasserman, G., & Allen, R. (1985). Maternal withdrawal from handicapped toddlers*. Journal of Child Psychology and Psychiatry, 26,* 381–387.

Wasserman, G.A., Allen, R., & Linares, L.O. (1988). Maternal interaction and language development in children with and without speech-related anomalies. *Journal of Communication Disorders, 21,* 319–331.

Wasserman, G.A., Allen, R., & Solomon, C.R. (1985). At-risk toddlers and their mothers: the special case of physical handicap. *Child Development, 56,* 73–83.

Wasserman, G., Lennon, M., Allen, R., & Shilansky, M. (1987). Contributors to attachment in normal physically handicapped infants. *Journal of American Academy of Child and Adolescent Psychiatry, 26*, 9–15.

Wehby, G. & Cassell, C. H. (2010). The impact of orofacial clefts on quality of life and healthcare use and costs. *Oral Diseases, 16*, 3–10.

Wehby, G. L. & Murray, J. C. (2010). Folic acid and orofacial clefts: a review of the evidence. *Oral Diseases, 16*, 11–19.

Wehby, G.L., Ohsfeldt, R.L., & Murray, J.C. (2006). Health professionals' assessment of health-related quality of life values for oral clefting by age using a visual analogue scale method. *The Cleft Palate-Craniofacial Journal, 43*, 383–391.

Weigl, V., Rudolph, M., Eysholdt, U., & Rosanowski, F. (2005). Anxiety, depression, and quality of life in mothers of children with cleft lip/palate. *Folia Phoniatrica et Logopaedica*, *57*, 20-27.

Weinberg, K., & Tronick, E.Z (1994). Beyond the face: An empirical study of infant affective configurations of facial, vocal, gestural and regulatory behaviors. *Child Development*, *65,* 1503-1515.

Weissman, M., Pilowsky, D.J., Wickramaratne, P.J., Talati, A., Wisniewski, S.R., Fava, M. et al. for the STAR*D-Child Team. (2006). Remissions in maternal depression and child psychopathology: A STAR*D-child report. *Journal of the American Medical Association, 295* (12), 1389–1398.

Wellens, W., & Vander-Poorten, V. (2006). Keys to a successful cleft lip and palate team. *B-ENT, 4,* 3–10. Available in https://www.ncbi.nlm.nih.gov/pubmed/17366839

Wheaton, B. (2007). The twain meets: distress, disorder and the continuing conundrum of categories. *Health, 11*, 303-319.

Whisman, M. A., Uebelacker, L. A., & Weinstock, L. M. (2004). Psychopathology and marital satisfaction: The importance of evaluating both partners. *Journal of Consulting and Clinical Psychology, 72* (5), 830-838.

Wijnberg-Williams, B.J., Van de Wiel, H.B.M., Kamps, W., & Hoekstra-Weebers, J.E.H.M. (2015). Effects of communication styles on marital satisfaction and distress of parents of pediatric cancer patients: a prospective longitudinal study. *PsychoOncology, 24,* 106–112. doi: 10.1002/pon.3617

Wilson, S., & Durbin, C.E. (2010). Effects of paternal depression on fathers' parenting behaviors: a meta-analytic review. *Clinical Psychology Review, 30*, 167– 180.

Winnicott, D.W. (1941). Observation of infants in a set situation. *International Journal of Psychoanalysis*, *22,* 229–249.

Wood, P. R., Hidalgo, H. A., Prihoda, T. J., & Kromer, M. E. (1993). Hispanic children with asthma morbidity. *Pediatrics*, *91*, 62–69.

Yazdy, M.M., Honein M.A., Rasmussen, S., & Frias, J.L. (2007a). Priorities for future public health research in orofacial clefts. *Cleft Palate Craniofacial Journal, 44,* 351–357. Available in http://www.cpcjournal.org/doi/pdf/10.1597/06-233.1

Yazdy, M., Honein, M. A. & Xing, J. (2007b). Reduction in orofacial clefts following folic acid fortification of the, U. S. grain supply. *Birth Defects Research. Part A Clinical and Molecular Teratology, 79,* 16–23.

Yeh, C.H. (2002). Gender differences of parental distress in children with cancer. *Journal Advanced Nursing, 38* (6), 598–606. doi: 10.1046/j.1365-2648.2000.02227.x

Yttri, J. E., Christensen, K., Knudsen. L. & Bille, C. (2011). Reproductive patterns among Danish women with oral clefts. *Cleft Palate-Craniofacial Journal, 48 (5), 601-607.* doi: 10.1597/09-245

Zahn-Waxler, C., Mayfield, A., Radke-Yarrow, M. R., McKnew, D. H., Cytryn, L., & Davenport, Y. B. (1988). A follow-up investigation of offspring of parents with bipolar disorder. *American Journal of Psychiatry, 145,* 506–509.

Zeanah, C.H., Boris, N.W., Heller, S.S., Hinshaw-Fuselier, S., Larrieu, J.A., Lewis, M., Palomino, R., Rovaris, M., & Valliere, J. (1997). Relationship assessment in infant mental health. *Infant Mental Health Journal, 18,* 182–197.

Zeytinoglu, S., & Davey, M.P. (2012). It's a privilege to smile: impact of cleft lip and palate on families. *Families, Systems, & Health, 30* (3), 265-277.

Zeytinoglu, S., Davey, M.P., Crerand, C., & Fisher, K. (2016a). Fathers of children born with cleft lip and palate: Impact of the timing of diagnosis. *Families, systems and Health, 34* (2), 150-158.

Zeytinoglu, S., Davey, M.P., Crerand, C., Fisher, K. & Akyil, Y. (2016b). Experiences of couples caring for a child born with cleft lip and/or palate: Impact of the timing of diagnosis. *Journal of Marital and Family Therapy, 43,* 82-99.

Zvara, B. J., Schoppe-Sullivan, S. J., & Dush, C. M. (2013). Fathers' involvement in child health care: Associations with prenatal involvement, parents' beliefs, and maternal gatekeeping. *Family Relations, 62,* 649–661. http://dx.doi.org/10.1111/fare.12023

6. APPENDIX

6.1. Glossary

- CL/P: Cleft Lip with or without Cleft Palate malformation.

- CLP: Cleft Lip and Palate malformation.

- CL: Cleft Lip malformation.

- ADBB: Alarm Distress Baby Scale.

- M-ADBB: Modified version of the Alarm Distress Baby Scale.

- PIPE: Pediatric Infant Parent Exam.

- IOFS: Impact On Family Scale.

- IDP: Psychological Distress Index.

- EPDS: Edinburgh Postnatal Depression Scale.

- DAS: Dyadic Adjustment Scale.

6.2. Informed consent

FORMULAIRE DE CONSENTEMENT

Le Professeur / Docteur [1] (Nom)_____ (Prénom)_____, médecin investigateur, nous a proposé de participer avec notre enfant à la recherche intitulée :

« Développement relationnel des enfants porteurs de fentes labio-palatines : influence du délai précédant la première intervention chirurgicale et de la perception psychologique de l'anomalie par les parents»

- Après avoir reçu oralement et par écrit toutes les informations nécessaires pour comprendre l'intérêt de cette recherche, ses contraintes et ses risques éventuels, ainsi que ce qu'il nous sera demandé de faire dans le cadre de notre participation,
- Après avoir pu poser les questions que nous souhaitions à propos de cette recherche et de ses implications sur notre prise en charge et obtenu des réponses,
- Après avoir bénéficié d'un délai de réflexion suffisant,
- Nous savons que nous pouvons retirer à tout moment notre consentement à notre participation à cette recherche, et cela quelles que soient nos raisons et sans supporter aucune responsabilité. Le fait de ne plus participer à cette recherche ne portera pas atteinte à nos relations avec le médecin investigateur qui nous proposera, si besoin, une autre prise en charge.
- Le Comité de Protection des Personnes Est IV a émis un avis favorable à la mise en œuvre de cette recherche le _____et l'Autorité Compétente a donné son autorisation le 12/10/2009.
- Les données personnelles recueillies au cours de cette recherche pourront être transmises, dans le respect du secret professionnel, au représentant du promoteur de la recherche et des autorités de santé, dans un but de contrôle de conformité.
- Un traitement de nos données personnelles va être mis en œuvre pour permettre d'analyser les résultats de la recherche au regard de l'objectif de cette dernière qui nous a été présentée. A cette fin, les données médicales nous concernant et les données relatives à nos habitudes de vie seront transmises au promoteur de la recherche ou aux personnes ou sociétés agissant pour son compte, en France ou à l'étranger. Ces données seront identifiées par un numéro de code correspondant aux trois premières lettres du nom et prénom de notre enfant. Conformément aux dispositions de la loi relative à l'informatique, aux fichiers et aux libertés, nous disposons également d'un droit d'opposition à la transmission des données couvertes par le secret professionnel susceptibles d'être utilisées dans le cadre de cette recherche et d'être traitées.
- Nous pouvons également accéder, directement ou par l'intermédiaire d'un médecin de notre choix, à l'ensemble de nos données médicales, en application des dispositions de l'article L1111-7 du Code de la Santé Publique. Ces droits s'exercent auprès du médecin qui nous suit dans le cadre de la recherche et qui connaît notre identité.
- Les Hôpitaux Universitaires de Strasbourg, promoteur de cette recherche, ont contracté une assurance, conformément à la loi.
- Nous sommes affiliés à un régime de Sécurité Sociale.
- Votre enfant sera filmé pendant 10 à 15 minutes lors des deux entretiens prévus avec la psychologue.

- *Une indemnité me sera versée pour mes frais de déplacements*

Après en avoir discuté et avoir obtenu une réponse à toutes nos questions, nous acceptons librement et volontairement de participer à cette recherche et **nous donnons notre accord pour y participer.**

Notre signature atteste que nous avons clairement compris les renseignements concernant notre participation à ce projet de recherche.

Enfant	Nom :	Prénom :

Investigateur :	**Personnes donnant le consentement :**	
	Père	Mère
Nom :	Nom :	Nom :
Prénom :	Prénom :	Prénom :
Date :	Date :	Date :
Signature :	Signature :	Signature :

(1) rayer la (les) mention(s) inutile(s)

Feuillet blanc à conserver 15 ans par l'investigateur – Feuillet jaune à conserver par le patient

6.3. Instruments

6.3.1. Alarm Distress Baby Scale (ADBB) (Guedeney & Fermanian, 2001)

ALARME DETRESSE BEBE (ADBB)

DATE : **PERIODE : T0 T1**

AGE : |__|__| mois |__|__| jours **Examinateur :**

Chaque item est coté de 0 à 4.

0 : Pas de comportement anormal de retrait
1 : Comportement discrètement anormal
2 : Comportement nettement anormal
3 : Comportement très nettement anormal
4 : Comportement massivement anormal

L'échelle est au mieux remplie par l'observateur lui-même, sur la base de ses propres observations, juste après la consultation. On évalue d'abord le comportement spontané, puis la réaction aux stimulations (sourire, voix, geste, toucher, etc..), en suivant l'évolution des réactions tout au long de l'examen. La valeur correspond à la réaction la plus significative pendant toute la durée de l'observation. En cas d'hésitation entre deux valeurs de l'échelle, on applique la gradation ci-dessus. **En cas de doute, on applique la valeur la plus basse.**

1 EXPRESSION DU VISAGE. Diminution de l'expressivité du visage:

0 : Le visage est spontanément mobile, expressif, animé par de fréquents changements d'expression.
1 : Visage mobile, expressif, mais sans changements fréquents d'expression.
2 : Peu de mobilité faciale spontanée.
3 : Visage immobile.
4 : Visage figé, froid, absent.

2 CONTACT VISUEL. Diminution du contact visuel:

0 : Contact visuel spontané facile et prolongé.
1 : Contact visuel spontané, mais bref.
2 : Contact visuel possible, mais seulement lorsqu'il est recherché.
3 : Contact visuel fugace, vague, fuyant.
4 : Evitement total du contact visuel.

3 ACTIVITE CORPORELLE. : Diminution de la mobilité de la tête, du torse et des membres, sans prendre en compte l'activité des mains et des doigts :

0 : Mouvements fréquents et spontanés du torse, de la tête et des membres.
1 : Activité générale spontanée légèrement réduite, peu d'activité de la tête ou des membres.
2 : Peu ou pas d'activité spontanée, mais activité présente en réponse à la stimulation.
3 : Faible activité en réponse à la stimulation.
4 : Enfant immobile et figé, quelle que soit la stimulation.

4 GESTES D'AUTO-STIMULATION. L'enfant se centre son corps (doigts, mains, cheveux, succion du pouce, frottement répétitifs...), d'une manière automatique, sans plaisir, et de façon apparaissant détachée du reste de son activité' :

0 : Absence d'autostimulation, l'activité d'auto exploration est en rapport harmonieux avec le niveau d'activité général
1 : Autostimulation fugitive
2 : Autostimulation peu fréquente mais nette
3 : Autostimulation fréquente
4 : Autostimulation constante ou quasi constante **Suite au verso**

5 VOCALISATIONS. Diminution des vocalisations, qu' elles traduisent le plaisir (gazouillis, rire, babil, lallations, cris aigus de plaisir), mais aussi le déplaisir, l'anxiété ou la douleur (cris, geignements et pleurs) :

0 : Vocalisations positives spontanées fréquentes, plutôt gaies et modulées ; cris ou pleurs brefs en réponse à une sensation désagréable.
1 : Vocalisations spontanées positives brèves
2 : **Vocalisations spontanées rares.**
3 : **Geignement** en en réponse à une stimulation.
4 : Aucune vocalisation**, même en cas de stimulation nociceptive.**

6 VIVACITE DE LA REACTION A LA STIMULATION. Diminution de la vivacité de la réaction à la stimulation, au cours de l'examen (sourire, voix, toucher). Note: ce n'est pas l'ampleur de la réponse qui est évaluée ici, mais le délai de la réponse **; l'absence de réaction ne permet pas de coter:**

0 : Réaction adaptée, vive et rapide.
1 : Réaction légèrement retardée.
2 : Réaction nettement retardée.
3 : Réaction nettement retardée, même en réponse à une stimulation désagréable.
4 : Réaction très retardée.

7 RELATION. Diminution de l'aptitude de l'enfant à entrer en relation, avec l'observateur, l'examinateur ou toute personne présente dans la pièce, exceptée celle qui s'occupe habituellement de l'enfant. La relation est évaluée par le comportement, le contact visuel, la réaction aux stimulations :

0 : La relation rapidement et nettement établie reste soutenue (après une éventuelle phase initiale d'anxiété).
1 : Relation identifiable, positive, ou négative, mais moins marquée ou soutenue qu'en 0
2 : Relation peu marquée, positive ou négative, peu soutenue.
3 : Relation à peine marquée.
4 : Absence de relation identifiable à l'autre.

8 - ATTRACTIVITE. Effort d'attention nécessaire pour rester en contact avec l'enfant, et sentiment de plaisir ou d'inquiétude que procure le contact avec l'enfant, **et le sentiment subjectif de durée de l'examen :**

0 : L'enfant attire l'attention par ses initiatives, sans aucun effort tout au long de l'examen, et inspire un sentiment d'intérêt et de plaisir, sans aucune inquiétude.
1 : Pas d'inquiétude, mais sentiment d'attraction moins marqué et soutenu.
2 : Sentiment neutre vis-à-vis de l'enfant, avec parfois du mal à garder durablement son attention centrée sur lui.
3 : Sentiment de malaise, d'être maintenu à distance, inquiétude nette.
4 : Contact éprouvant, sentiment d'un enfant hors d'atteinte, très préoccupant.

Total :

6.3.2. Modified version-Alarm Distress Baby Scale (m-ADBB) (Matthey, Crncec, Hales, & Guedeney, 2013)

Echelle M-ADBB

Chaque item est coté selon les catégories suivantes :

0 : Satisfaisant
1 : Problème possible
2 : Problème manifeste

Cette échelle est au mieux remplie par l'observateur, sur la base des ses observations lors de l'examen clinique. Le clinicien devrait essayer d'interagir avec l'enfant, par le sourire, la parole ou le contact physique.

1. EXPRESSION FACIALE :
L'observateur évalue le degré de l'expressivité facile pendant l'examen :

0 : Satisfaisant :
Le visage montre une expressivité nette, qu'elle soit positive (sourire) ou négative grimace)

1: Problème possible :
L'expressivité du visage est limitée, mais elle existe, de façon positive ou négative.

2 : Problème manifeste :
Pas d'expression faciale ; le visage apparaît rigide, immobile, ou triste pendant toute la période d'observation.

2. CONTACT OCULAIRE :
L'observateur évalue la qualité du contact oculaire, avec lui-même ou elle-même :

0 : Satisfaisant :
Au moins un épisode de contact de durée moyenne, avec plusieurs épisodes de contact bref.

1 : Problème possible :
Seulement deux épisodes de contact oculaire, ou seulement un épisode de durée moyenne.

2 : Problème manifeste :
Un seul contact bref, ou un contact oculaire vague, évitant, ou complètement absent.

3. VOCALISATIONS, EVALUEES VIS A VIS DE TOUS
L'observateur évalue la quantité de vocalisation exprimant le plaisir (gazouillis, rire, babil, lallations, cris aigus de plaisir), mais aussi le manque de vocalisation exprimant le déplaisir ou la douleur (cris ou pleurs), tout au long de l'observation).

0 : Satisfaisant :
Au moins quelques vocalisations brèves (sans pleurs), ou une ou deux longues.

1: Problème possible :
Seulement quelques vocalisations sans pleurs, ou si aucune de ces vocalisations, au moins des cris ou des pleurs en réponse à la stimulation, ou des geignements nets.

2 : Problème manifeste :
L'enfant ne geint qu'occasionnellement, et seulement en réponse à la stimulation, ou bien absence totale de vocalisation.

4. ACTIVITE, EVALUEE VIS A VIS DE TOUS

On évalue la fréquence des mouvements de l'enfant au niveau due la tête, du torse et des membres, spontanément et en réponse à la stimulation, agréable ou désagréable, mais sans prendre en compte l'activité des mains et des doigts.

1 : Satisfaisant :
Un niveau au moins modéré d'activité, avec quelques mouvements de la tête, du torse et des membres.

2 : Problème possible :
Niveau d'activité spontané très réduit, peu de mouvements de la tête, des membres et du torse, seulement en réponse à la stimulation.

3: Problème manifeste :
pas d'activité spontanée, ou très faible niveau d'activité en réponse à la stimulation.

5. RELATION : VIS-A-VIS DU CLINICIENOU DE L'OBSERVATEUR SEULEMENT :

L'observateur évalue la capacité de l'enfant à rentrer en relation avec lui ou elle, ou avec toute autre personne présente, à l'exception des parents. La capacité d 'entrer en relation est évaluée au travers de l'attitude de l'enfant vis à vis de l'autre, du contact visuel, et des sa réaction à la stimulation et dans l'interaction

0 : Satisfaisant :
La relation, positive ou négative, est au moins présente de façon modérée.

1 : Problème possible :
La relation est présente de façon tenue, ou douteuse, ou seulement quand l'enfant pleure, ou se débat.

2 : Problème manifeste :
pas de relation manifeste, que ce soit sur un mode positif ou négatif.

Caractéristiques du clinicien dans l'examen :

1. Tente activement d'engager l'enfant dans la relation (sourires, parle à l'enfant)
2. Tente effectivement d'engager l'enfant dans la relation (sourires, parle à l'enfant)
3. Tente peu d'engager l'enfant dans la relation (sourires, parle à l'enfant).

Caractéristiques de l'enfant pouvant influer sur l'évaluation :

1. L'enfant apparaît fatigué
2. L'enfant apparaît en détresse pendant tout l'examen
3. Autre (préciser)

Nombre de satisfaisant: ___; Nb de problème possible : ____; Nb de problème manifeste: ___

DATE: AGE: NOM:

EXAMINATEUR: LIEU:

(Dérivée de la forme complète de l'échelle ADBB: Guedeney & Fermanian, 2001).

6.3.3. Pediatric Infant Parent Exam (PIPE) (Fiese, Poehlmann, Irwin, Gordon, & Curry-Bleggi, 2001)

PEDIATRIC INFANT PARENT EXAM (PIPE)

FIESE B., POEHLMANN J., IRWIN M., GORDON M., CURRY-BLEGGI E. A pediatric screening instrument to detect problematic infant-parent interactions: initial reliability and validity in a sample of high-and low-risk infants.

LE DEMARRAGE DU JEU

0. Engagement facile. Le parent attire facilement l'attention de l'enfant et l'enfant montre les affects positifs (le visage de l'enfant s'anime au son de la voix parentale ; le parent caresse l'enfant).
1. L'enfant a difficulté à s'engager. Le parent a du mal à capter l'attention de l'enfant mais l'enfant présente des affects positifs ou neutres (le parent fait sautiller l'enfant à plusieurs reprises, l'enfant finit par regarder ou sourire).
2. Le parent ne s'engage pas. Le parent n'est pas tout à fait présent mais l'enfant reste positif ou neutre (le parent jette constamment des regards vers le pédiatre ; l'enfant regarde le parent avec interrogation).
3. Le parent ne s'engage pas / l'enfant proteste. Le parent s'empresse peu à capter l'attention de l'enfant puis l'enfant proteste doucement ou regarde dans le vide.
4. Engagement intrusif / l'enfant évite ou est désintéressé. Le parent tapote à plusieurs reprises l'enfant pour attirer son attention et l'enfant tourne la tête ailleurs, proteste ou est inexpressif (le parent tapote l'enfant et continue même quand l'enfant tourne la tête ailleurs ou pleurniche).
5. Engagement inapproprié ou bizarre. Le parent commence le jeu avant que l'enfant ne le regarde ou peut dire des choses bizarres sans rapport avec le jeu. L'enfant dévisage le parent ou semble être détaché (le parent commence le jeu de manière bizarre, l'enfant regarde autour).

L'ENTRETIEN DU JEU

0. Les parents et l'enfant prennent du plaisir à jouer ensemble. Mouvements calmes, doux d'avant en arrière entre les parents et l'enfant, l'enfant peut rire ou sourire (le parent chantonne, s'arrête, l'enfant rit, le parent répète).
1. L'enfant a du mal à poursuivre le jeu. Le parent a du mal à capter l'attention de l'enfant mais l'enfant reste avec des affects positifs ou neutres (le parent augmente le son de sa voix - son activité physique pour maintenir l'attention de l'enfant ; l'enfant regarde le parent sans le regarder dans les yeux).
2. Le parent est détaché. Le parent joue de façon mécanique mais l'enfant reste neutre (peu ou pas d'attention au regard de l'enfant, peu de variation dans l'intensité et la tonalité de la voix, peu de variation dans le toucher ; l'enfant peut sembler

méfiant en fronçant les sourcils mais reste avec des affects neutres ou positifs).

3. Le parent est détaché. Le parent joue de façon mécanique, l'enfant peut s'agiter-pleurnicher (le parent montre peu ou pas de variation dans sa voix, dans le toucher ; l'enfant détourne son regard ou semble s'agiter).
4. Stimulation intrusive / l'enfant évite ou reste passif. Stimulations physiques importantes en touchant poussant l'enfant ; l'enfant regarde ailleurs ou reste mou (le parent effectue des mouvements saccadés avec peu ou pas d'affects positifs. L'enfant se renferme ou proteste).
5. La façon de jouer est inappropriée. Le parent joue sans se soucier de l'enfant, l'enfant a un regard vide ou pleure (le parent parle du jeu ou d'un sujet sans rapport avec le jeu pendant qu'il joue, ne réponds pas aux protestations de l'enfant ; l'enfant peut fixer du regard ou pleurer).

L'ARRET DU JEU

0. Le jeu s'arrête progressivement. Le parent et l'enfant s'arrêtent tous deux de jouer progressivement (le parent parle moins fort ; l'enfant gazouille, redevient plus tranquille).
1. L'arrêt du jeu est en décalage. Le parent met fin au jeu, l'enfant proteste légèrement mais rapidement s'apaise (le parent arrête le jeu devant des signaux de l'enfant ou peut continuer d'apaiser l'enfant quand le jeu est terminé ; l'enfant proteste un peu et revient à un affect positif rapidement).
2. Arrêt brusque. Le parent arrête le jeu brutalement et l'enfant reste neutre (le parent peut faire une déclaration au pédiatre « ça suffit » ; l'enfant peut rester neutre ou se renferme).
3. Arrêt brusque / l'enfant proteste. Le parent arrête brutalement le jeu et l'enfant proteste (le parent arrête le jeu avec peu d'attention à l'enfant ; l'enfant proteste, le parent ne l'apaise pas).
4. L'intrusion parentale persiste. Quand le jeu est terminé, le parent continue de tapoter l'enfant, l'enfant reste figé ou proteste (le parent arrête de jouer mais continue d'exciter l'enfant ; l'enfant s'agite, se détourne du parent ou ne réponds pas).
5. Incapacité à arrêter le jeu. Le parent continue de jouer alors que l'enfant pleure ou n'exprime pas d'affect (le parent continu de jouer même quand le pédiatre parle).

Total :

6.3.4. Impact On Family Scale (IOFS) (Boudas, Jégu, Grollemund, Quentel, Danion-Grilliat, & Velten, 2013)

IOFS

Pour chaque énoncé ci-dessous, veuillez indiquer (en encerclant le chiffre correspondant) si vous êtes tout à fait d'accord, d'accord, pas d'accord ou pas du tout d'accord.

		Tout à fait d'accord	D'accord	Pas d'accord	Pas du tout d'accord
1	À cause de la maladie de notre enfant nous ne pouvons pas faire de grands trajets.	1	2	3	4
2	Les gens du voisinage se comportent différemment avec nous à cause de la maladie de mon enfant.	1	2	3	4
3	Nous avons peu envie de sortir à cause de la maladie de mon enfant.	1	2	3	4
4	Il est difficile de trouver une personne de confiance pour prendre soin de mon enfant.	1	2	3	4
5	Nous devons parfois modifier nos projets de sortie à la dernière minute en raison de l'état de notre enfant.	1	2	3	4
6	Nous voyons moins souvent notre famille et nos amis, à cause de la maladie de notre enfant.	1	2	3	4
7	Parfois je me demande s'il faudrait se comporter avec mon enfant de manière particulière ou de la même façon qu'avec un autre enfant.	1	2	3	4
8	Je ne pense pas avoir d'autres enfants, à cause de la maladie de mon enfant.	1	2	3	4
9	Il ne me reste pas beaucoup de temps à consacrer aux autres membres de ma famille une fois que j'ai prodigué les soins nécessaires à mon enfant.	1	2	3	4
10	Notre famille renonce à certaines choses à cause de la maladie de notre enfant.	1	2	3	4
11	Je suis souvent fatigué(e) à cause de la maladie de mon enfant.	1	2	3	4
12	Je vis au jour le jour et je ne fais pas de projets pour l'avenir.	1	2	3	4
13	Personne ne comprend ce que je dois supporter.	1	2	3	4
14	Les déplacements à l'hôpital me pèsent.	1	2	3	4
15	Parfois, j'ai l'impression que ma vie ressemble aux montagnes russes : très difficile quand mon enfant va mal, sans difficulté particulière quand il va bien.	1	2	3	4

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Adapté pour la France par Raphaël Boudas, Michel Velten, Bruno Grollemund, Elvire Quentel.

6.3.5. Psychological Distress Index (IDP) (Préville, Boyer, Potvin, Perrault, & Légaré, 1992)

IDP **Version courte**

Facteur anxiété :

1- Vous êtes-vous senti(e) tendu(e) ou sous pression ?	OUI O NON O
2- Avez-vous ressenti des peurs ou des craintes ?	OUI O NON O
3- Vous êtes-vous senti(e)agité(e) ou nerveux (se) intérieurement ?	OUI O NON O

Facteur dépressif :

4- Vous êtes-vous senti(e) désespéré(e) en pensant à l'avenir ?	OUI O NON O
5- Vous êtes-vous senti(e) seul(e) ?	OUI O NON O
6- Vous êtes-vous senti(e) découragé(e) ou avez-vous eu le blues ?	OUI O NON O
7- Vous êtes-vous senti(e) ennuyé(e) ou peu intéressé(e) par les choses?	OUI O NON O
8- Avez-vous pleuré facilement ou vous êtes-vous senti(e) sur le point de pleurer ?	OUI O NON O

Facteur irritabilité :

9- Vous êtes vous laissé(e) emporté(e) contre quelqu'un ou quelque chose ?	OUI O NON O
10- Vous êtes-vous senti(e) négatif (ve) envers les autres ?	OUI O NON O
11- Vous êtes-vous senti(e) facilement contrarié(e) ou irrité(e) ?	OUI O NON O
12- Vous êtes-vous fâché(e) pour des choses sans importance ?	OUI O NON O

Facteur problèmes cognitifs :

13- Avez-vous eu des blancs de mémoire ?	OUI O NON O
14- Avez-vous eu des difficultés à vous souvenir des choses ?	OUI O NON O

6.3.6. Edinburgh Postnatal Depression Scale (EPDS) (Guedeney & Fermanian, 1998)

EDPS

Vous venez d'avoir un bébé. Nous aimerions savoir comment vous vous sentez. Nous vous demandons de bien vouloir remplir ce questionnaire en cochant la réponse qui vous semble la mieux décrire comment vous vous êtes sentie durant la semaine (c'est-à-dire sur les 7 jours qui viennent de s'écouler) et pas seulement aujourd'hui.

Pendant la semaine qui vient de s'écouler :

1- J'ai pu rire et prendre les choses du bon côté.

- ❑ Aussi souvent que d'habitude
- ❑ Pas tout à fait autant
- ❑ Vraiment beaucoup moins souvent ces jours-ci
- ❑ Absolument pas

2- Je me suis senti(e) confiant(e) et joyeux (se), en pensant à l'avenir.

- ❑ Autant que d'habitude
- ❑ Plutôt moins que d'habitude
- ❑ Vraiment moins que d'habitude
- ❑ Pratiquement pas

3- Je me suis reproché, sans raison, d'être responsable quand les choses allaient mal.

- ❑ Oui, la plupart du temps
- ❑ Oui, parfois
- ❑ Pas très souvent
- ❑ Non, jamais

4- Je me suis senti(e) inquiet(e) ou soucieux (se) sans motifs.

- ❑ Non, pas du tout
- ❑ Presque jamais
- ❑ Oui, parfois
- ❑ Oui, très souvent

5- Je me suis senti(e) effrayé(e) ou paniqué(e) sans vraiment de raisons.

- ❑ Oui, vraiment souvent
- ❑ Oui, parfois
- ❑ Non, pas très souvent
- ❑ Non, pas du tout

Merci de compléter la suite au verso →

6- J'ai eu tendance à me sentir dépassé(e) par les événements.

- ❑ Oui, la plupart du temps, je me suis sentie incapable de faire face aux situations
- ❑ Oui, parfois, je ne me suis pas sentie aussi capable de faire face que d'habitude
- ❑ Non, j'ai pu faire face à la plupart des situations
- ❑ Non, je me suis sentie aussi efficace que d'habitude

7- Je me suis senti(e) si malheureux(se) que j'ai eu des problèmes de sommeil.

- ❑ Oui, la plupart du temps
- ❑ Oui, parfois
- ❑ Pas très souvent
- ❑ Non, pas du tout

8- Je me suis senti(e) triste ou peu heureuse.

- ❑ Oui, la plupart du temps
- ❑ Oui, très souvent
- ❑ Pas très souvent
- ❑ Non, pas du tout

9- Je me suis senti(e) si malheureux(se) que j'en ai pleuré.

- ❑ Oui, la plupart du temps
- ❑ Oui, très souvent
- ❑ Seulement de temps en temps
- ❑ Non, jamais

10- Il m'est arrivé de penser à me faire du mal.

- ❑ Oui, très souvent
- ❑ Parfois
- ❑ Presque jamais
- ❑ Jamais

6.3.7. Dyadic Adjustment Scale (DAS) (Antoine, Christophe, & Nandrino, 2008)

Echelle de Spanier

Veuillez indiquer votre réponse en cochant la case qui correspond à ce que vous vivez.

Nous sommes généralement d'accord dans les domaines suivants	Jamais d'accord	Rarement d'accord	Parfois d'accord	Assez souvent d'accord	La plupart du temps d'accord	Toujours d'accord
01 Les objectifs, les buts et ce qu'on trouve important dans la vie						
02 Les prises de décision importantes						
03 Les marques d'affection						
04 Les amis						
05 Les relations sexuelles						
06 La philosophie de la vie						
07 Les façons d'agir avec les parents et les beaux parents						

Pour chacune des phrases suivantes, veuillez indiquer votre réponse en cochant la proposition qui correspond à ce que vous vivez	Jamais	Rarement	Parfois	Assez souvent	La plupart du temps	Toujours
08 Il m'arrive de penser au divorce, à la séparation ou à terminer notre relation						
09 Nous nous « tapons sur les nerfs »						
10 Nous avons des échanges d'idées stimulants						
11 Nous discutons calmement						
12 Je me confie à mon partenaire						
13 Nous avons des intérêts communs à l'extérieur de la maison						
14 Nous rions ensemble						
15 Nous travaillons ensemble à un projet						

	Extrêmement malheureux	Passablement malheureux	Un peu malheureux	Heureux	Très heureux	Extrêmement heureux
16 Quel est globalement votre degré de bonheur dans votre relation?						