The relational development of infants with CLP: impact on parents and on the parent-infant relationship. Results from the first French prospective multicenter CLP study.

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Abstract

Background: The objective of this prospective, multidisciplinary and multicenter study was to explore the impact of a cleft lip, whether or not in association with a cleft palate (CLP) on the parents, on the parent-infant relationship, and on the infant's relational development, depending on the type of malformation and the timing of the repair surgery, at 4 and 12 months postpartum. Method: 158 infants with CLP and their parents participated in this multicenter prospective cohort. Clinical evaluations were performed at 4 and 12 months postpartum. The impact on the parents and on the parent-infant relationship was evaluated by the Parenting Stress Index (PSI), the Edinburgh Post-partum Depression Scale (EPDS) and the Impact-on-Family Scale (IOFS). The relational development of the infant was assessed using the Alarm Distress Baby Scale (ADBB). The main criteria used to compare the infants were the severity of CLP and the time of surgery. Results 1) No effect was found in this sample of the timing of surgery, the type of malformation or the care structure on social withdrawal behaviors of the child at 4 and 12 months postpartum (ADBB). Furthermore, early intervention significantly decreased maternal stress assessed with the PSI at 4 months. 2) Parents for whom it had been possible to give a prenatal diagnosis were much better prepared to accept the waiting time between birth and the first surgical intervention (PSI, EPDS, IOFS). 3) Higher postpartum depression scores (EPDS) were found for both parents compared to the general population. Conclusion: A joint assessment of the mental health of both infants and parents is required in the follow-up of cleft lip and palate. Even if most families are remarkably resilient when faced with this major cause of stress, a significant proportion of them could require help to deal with the situation, especially during this first year of follow-up. An assessment of social withdrawal behaviours in the child and of the level of parental stress and depression appears useful, in order to adapt care to infant and parents alike. Trial Registration: ClinicalTrials.gov Identifier: NCT00993993. Registered 10/14/2009, https://clinicaltrials.gov/ct2/show/NCT00993993?term=grollemund&draw=2&rank=1.

Background

Cleft Lip and Palate (CLP) is the most frequent congenital craniofacial malformation in humans (1). Three different types of CLP are distinguished: Cleft Lip, Cleft Lip and Palate, and Cleft Palate only. This medical condition is not a major cause of mortality in developed countries; however, it does cause considerable morbidity among children who are affected, and imposes a substantial financial load for families, with an associated societal burden (2).

Previous studies have observed a higher prevalence of psychiatric comorbidities for children with CLP. Recently, Tillman et al. (3) found that patients with CLP showed an increased prevalence of psychiatric illness, intellectual disability, language disorders, psychotic disorders, Autism Spectrum Disorders (ASD), attention deficit/hyperactivity disorders, and other behavioural disorders beginning in childhood. In addition, difficulties in interactional skills among subjects with CLP have been observed (4). Relational difficulties for infants with CPL have been observed in previous studies. For example, Habersaat et al. (5) found that infants with CLP were more difficult and less cooperative in interactions with their mothers at 2 months of age, compared to controls. Previous studies have indicated that the interaction style between
parents and infants with CLP differs significantly from that of control dyads (6). These authors observed that infants with a cleft lip were less implicated in the interaction, and their mothers were less responsive and less sensitive in the interpretation of their infant’s signals. This suggests that the detection of CLP with its visible facial disfigurement could complicate the transition to parenthood and the parents’ first encounter with their infant (7), so that the parent-infant relationship may form under stressful circumstances.

In addition, when CLP is diagnosed, parents are likely to experience emotional reactions such as confusion, denial, distress, and guilt (8), as well as feelings of loss of control, helplessness, and even depression and anxiety (9). Parents can feel damaged by their perceived inability to produce a healthy and typically formed baby who is free from any physical defects (10-12). As a result, parents have to go through a grieving process, which is necessary for their acceptance of the fact that their child is different (9). Consequently, the announcement of a facial malformation affecting their baby is a major psychological burden for parents.

In addition to this, the different types of malformation have a different impact on the parent-infant relationship. Since the CLP directly affects the infant’s face, the parents’ attachment to the baby can be affected (13). For example, in mother-infant face-to-face interactions, Endriga and Speltz (14) found that mothers of more mildly impaired infants (cleft palate only) were more distant than mothers of more severely impaired infants (cleft lip and palate). The authors suggested that these results could be explained firstly because mothers received less professional attention and support in case of cleft palate alone than those with infants with cleft lip and palate. On the other hand, Despars et al. (7) observed that the severity or complexity of the cleft was not related to parental representations and posttraumatic stress. These inconclusive findings led us to think it would be worthwhile evaluating the impact of CLP on the parent-infant relationship according to the type of CLP.

The treatment of CLP is highly complex, requiring extensive therapy to remediate speech, language, hearing, and feeding deficits. In most cases, numerous surgical procedures are required to correct the associated anatomical deficits (15). Currently, the modalities of reconstructive surgery vary according to the severity of the malformation and the protocol followed by each surgical team. To date, no generic protocol has reached a consensus in the entire medical community; therefore, each patient is treated according to the experience and the decisions of the medical team in charge (16).

It has been suggested that the timing of surgical repair could affect parent-infant interactions in the context of cleft lip, because of the dramatic cosmetic change (17). Previous studies have suggested that early repair could promote a better parent-infant interaction, since early repair of the CLP could facilitate parents’ responses to social cues from the infant (8, 18). On the other hand, late repair could be advantageous, because parents have time to adjust to the infant’s condition, and plan the surgery along with the medical team (18).

Murray et al. (19) for their part observed that mothers of infants undergoing late repair surgery were less positively involved with their infant, and infants aged 2 months also spent less time looking at their
mother. Surprisingly, at six months and at twelve months, there were no differences in the mother-infant relationship between the groups (early repair, late repair, and controls). Nevertheless, in the same study, the mothers of more disfigured infants were less positively involved, and the infants with both cleft lip and palate looked less frequently at their mothers in the early and late repair groups. These findings support the fact that having a child with CLP can adversely affect the parent-infant relationship.

In this paper, we aimed to study social withdrawal evaluated with the ADBB scale in a prospective cohort of 156 infants with CLP. Our objectives were to identify factors associated with withdrawal by the infant at 12 months, and to describe parental stress and depression during the first year.

**Methods**

**Participants**

A detailed description of the inclusion criteria, the recruiting centers, and all items assessed can be found in Grollemund et al. (16).

**Instruments**

The different measures are thoroughly described in (16). Only the results pertaining to the main hypothesis are presented here.

1) Social withdrawal behaviour by the child, as assessed with the ADBB on video clips filmed during follow-up consultations with parent and infant

2) The Parenting Stress Index (PSI), which enables screening for parental attitudes that could be risk factors for the development of emotional and developmental disturbances in a young child.

3) The Edinburgh Post-partum Depression Scale (EPDS) completed by each parent, in its validated French version.

4) The Impact on Family Scale (IOFS) assessing the family, social and financial impact of the malformation.

**Design**

This study was a prospective cohort of children diagnosed with CLP. Two evaluation periods were planned: T0, when the infant was 4 months, and T1 when the child was 12 months, i.e. at least 6 months after the first surgical intervention. Only children with CLP were included, isolated or familial, either syndromic (associated with other abnormalities or malformations) or non-syndromic. Parents were included following informed consent for themselves and their child. The ADBB score measured at 12 months was used as the main criterion to identify factors associated with social withdrawal.

**Statistical procedures**
In the descriptive analysis, patient characteristics were described using frequencies and percentages for categorical variables, and medians and inter-quartile range values for continuous variables. Correlations between ADBB, PSI, IOFS and EPDS scores were estimated using Spearman's rank correlation coefficient. Baseline factors associated with the ADBB score at T1 among the clinical and demographic characteristics (type of center, time lapse to surgical intervention, diagnosis, side of the cleft, type of cleft, interaction between side and type of cleft) were identified using univariate linear models. The effect of each binary variable on the ADBB score was analysed by way of the differences of means and a 95% confidence interval was obtained by bootstrapping 10000 samples. We also used the p-value provided by the Wilcoxon non-parametric test. Factors associated with the PSI score were studied using the same methodology.

Sample size calculation: this study was initially powered to detect a difference of 2 ± 3.78 points for the ADBB score between groups with early and late surgical intervention at one year, with a power of 90%, an alpha error of 5% and assuming a dropout rate of 5%. Considering these hypotheses, the total study sample size calculated was 160 patients. This study was also performed to detect a difference of 22.9 ± 41.9 points for the PSI score between the 2 groups (early and late surgical intervention) using the same parameters. A p-value of <0.05 was considered statistically significant. Analyses were performed using SAS V.9.4 (SAS Institute Inc., Cary, North Carolina, USA) and R 3.4.0.

**Results**

Table 1 gives a description of the sample recruited: gender of the child, type of cleft, weeks’ gestation at the birth of the infant, mode of diagnosis (ante or postnatal), time lapse before first surgery (months), EPDS, PSI, and IOFS scores for mothers and fathers.

Table 2 shows the results of the univariate analysis for ADBB scores at T0 and T1

Table 3 shows the results of the univariate analysis for PSI maternal scores at T1

Figure 1 gives the study flowchart

Figure 2 gives a description of the evolution of ADBB scores between T0 and T1

Figures 3 shows a heat-map of correlations between instruments administered at T0 and T1

**Summary of the results**

- Two infants were excluded from the analysis in the antenatal diagnosis group because they lacked one of the two following criteria: age 0 to 4 months, or presence of unilateral or bilateral CLP. Only one child had a syndromic CLP, associated with other genetic abnormalities.

- In contrast with our main hypothesis that the longer the time-lapse before the first surgical intervention, the more likely the parents’ psychological perceptions would be to affect the harmonious development of
their child, there was no effect of the timing of the surgery on the social withdrawal behaviours of the child.

- There was no effect of the type of malformation on the level of social withdrawal behaviours at T0 (4 months), and no effect of the centre on the level of social withdrawal behaviours. The incidence of social withdrawal behaviours among CLP children (ADBB score >5) was 13%, the same level as that found in community studies in France.

- In line with our second hypothesis, parents who had been given a prenatal diagnosis were much better prepared to accept the waiting time between birth and the first surgical procedure.

- In line with our third hypothesis, negative feelings among parents in the later surgery group (3 to 6 months after birth) tended to decrease and to come into line with those of parents whose children had an early intervention. Alongside, the children’s distress tended to decrease from T0 to T1, as assessed with the Alarm Distress Baby scale (ADBB).

- Early intervention significantly decreased maternal stress as assessed with the PSI at 4 months.

- Both parents were affected by this experience and showed higher depression scores (EPDS) compared to the general population at T0 and T1, in the first months of treatment.

**Discussion**

Strengths of the study: We used a large array of instruments, first to assess social withdrawal behaviours among the infants using the well-validated ADBB scale, and secondly for both parents the EPDS, the PSI and the IOFS. We clearly distinguished the different forms of cleft, cleft lip with or without palate cleft. The assessment of social withdrawal behaviours was performed during routine follow-up consultations, using video clips. Independent scoring was conducted by an expert, and by the clinician immediately after the examination. A consensus score was calculated each time there was discrepancy between the scores given by the clinician in charge of the assessments and the expert (CPM).

Limitations: One limitation of the study was the large volume of missing data, especially for the secondary instruments (PSI, EPDS and IOFS). However, the availability of the ADBB score was good (only 10% missing data) and efforts were made to ensure a proper evaluation of this scale with a centralized evaluation based on video recordings. Other variables tested as potential risk factors for social withdrawal (severity of the cleft, time-lapse to the surgical procedure, prenatal diagnosis) had almost no missing data.

In addition, the results from the main analysis based on the ADBB were negative, which could suggest a lack of power of the study. Despite common representations, no association was found between social withdrawal evaluated with the ADBB scale and the severity of the cleft, and the reported mean difference was fairly small (i.e. clinically negligible). This result at least eliminates the likelihood of a strong
negative effect and highlights the fact that infants and parents should be followed and evaluated regardless of the severity of the malformation.

The time frame for the study was one year, which made it impossible to evaluate the potential negative impact of the cleft on language and development after one year of age.

Some parents refused to participate in the study, which potentially led to a selection bias in our sample. It can be thought that these refusals were mainly attributable firstly to the distance between home and the healthcare centre or the evaluation premises, and secondly to the repeated visits required for the child’s care, and also to some extent to the parents’ reluctance to confide their feelings and difficulties since the discovery of the malformation. As the study could not intrude on the private lives of these families, it was impossible to press for agreement, especially in cases where refusal came from one parent only. This applied more frequently to the father.

In the literature the parent–child relationship is always studied by interviewing the parents. The choice and the relevance of the instruments used can be questioned. Indeed, self-administered measures are subject to caution, as parental responses can lack objectivity. This is all the more so when data concerning families who refuse to participate is lacking. It is likely that this seriously biases the results obtained. Thus, it is difficult to assume that, overall, things are fairly satisfactory. Indeed, it would have been useful to know the number of families that did not take part in the study, and the reasons for their refusal, distinguishing personal reasons from practical reasons relating to travel or care centre organisation.

The families of children with CLP are not particularly likely to have experience of psychological or psychiatric support. They may be reluctant because of certain preconceived ideas about the speciality, and may argue that this consultation will not change reality and the problems they are facing, and above all they may not understand why interviews of this type could be useful. The assessment needs to be presented as an encounter enabling better acquaintance with the child, and better knowledge of any particular difficulties. In our encounters, we explained that if difficulties were identified, suitable care and support would be provided in a remediation process that is not solely physical.

It is very difficult for parents to express their feelings using the standardised questionnaires chosen. Certain items in the PSI were unsuited to the infants’ age in our study. However numerous parents took the opportunity provided by these interviews to talk with the psychologists and psychiatrists and confide their emotions. For some who felt particularly alone, this time for words undoubtedly provided assistance.

In the literature, the relationship between the parents and the child has always been studied only by questioning the parents, most often focusing solely on the mother. They answer the questions on the basis of their own feelings, which can only provide one aspect of reality. The ADBB scale enables the study of withdrawal behaviours in the child and focuses solely on the child. The scale has been used in a number of problematic neonatal situations, such as the Prader Willy syndrome (20), or neonatal cardiac surgery (21), but has never been used with children with CLP. Interestingly, the levels of social withdrawal
in these three situations (CLP, Prader-Willy, neonatal cardiac surgery) are not related to the severity of the medical condition, but rather to the level of stress and distress shown by the mothers of these infants. The main interest of this combined approach is that, for the first time, it puts the experience of the parents after a trauma into perspective, and evaluates its consequences on the child. The contribution of the ADBB scale for children in this age group in this instance enables a more objective study of the parent-child relationship, and in a more symmetrical manner. The behaviours of an infant, and any signs of withdrawal, are unlikely to be dissimulated, while this may not be the case with the parents.

Our findings emphasise the need to detect social withdrawal behaviours in the first months of life as a silent signal of suffering that should be interpreted and treated, especially when the infants concerned present a medical condition (21). Along these lines, Smith-Nielsen et al. (22) suggested that adding the ADBB to existing routine developmental health surveillance practices could add value to health care workers’ practice by improving their knowledge about the socio-emotional development of infants.

Given the importance of the relationship between parent and child, the clinical evaluation by surgeons should include screening of the mothers and fathers for symptoms of depression and anxiety at the time of their child's first evaluation. This would give the clinician an opportunity to engage parents in a dialogue about the relationship between their symptoms and their child's treatment outcomes. Even if they do not talk about it spontaneously, these "different" infants and their parents do suffer, and need someone to listen to their experiences, assessing their level of stress and possible difficulties relating to the situation. Indeed, it is important not to forget the many obstacles and hardships that these parents will have to overcome in this first year of life: accepting the fact that they have a child with a malformation, particularly for the mothers because this came about inside their body; at birth, seeing this cleft mouth for the first time; coping with the eyes of others; remaining creative and not collapsing as a parent when confronted with the refusals, failures and uncertainties of the first breast or bottle feeds; returning repeatedly to the hospital; coping with the anxiety of the first anesthesia; having to witness the child's discomfort after surgery; having to care for their baby; and once again accepting this baby after the changes brought about by surgery... The offer of an encounter with a psychologist or psychiatrist in a surgery department is one first essential aspect. But the way in which this is presented is also important. It should not be imposed, but it should be sufficiently advocated for any parent to readily take up the offer.

**Conclusion**

This study is the first in France, and to the best of our knowledge the first to address specifically the psychological consequences of the birth of a child with a CLP on the parents’ mental health and consequently on early parent-child relationships. In France, as elsewhere, the planning of the surgical interventions varies according to the care facility. Certain teams are in favour of early intervention, immediately after birth. Other teams prefer to wait three of even six months. This exploratory study provides teams with data on the effect of early surgery vs. later surgery. The resilience of both children and parents is remarkable, as the level of withdrawal behaviours in this exposed population was no
higher than that observed in the French community. On the other hand, given that the consequences of parental depression on infant development are well known, this study highlights parental needs in terms of psychological support, especially during the first year of treatment when the main procedures for closing the cleft take place. These results highlight the relevance of systematic screening for mental health issues among infants with cleft lip and palate and their parents.

Findings from a recent UK study suggest that the centralization of CL/P services has vastly enhanced patient experiences, and support the notion that psychologists should be integrated into each team, providing people with multidisciplinary professional care (23). This study was the first in France gathering the French expert centers and some of the more active centers around a common protocol to explore the psychological experiences of parents and children faced with this very demanding experience of a child born with a cleft lip and palate. The results of this study have already changed some of the professional practices in the French specialized centers, adding paediatric nurse appointments to assist with problems for the baby's feeding, to accompany the first mother-infant interactions, and to adjust the care to the infant and the parents.

**Abbreviations**

Cleft Lip malformation (CL); Cleft Lip with or without Cleft Palate malformation (CLP); the Alarm Distress baby scale (ADBB); the Parenting Stress Index (PSI); the Edinburgh Post-partum Depression Scale (EPDS); the Impact on Family Scale (IOFS);

**Declarations**

**Ethics approval and consent to participate**

This study was approved by the Comité de Protection des Personnes Est IV of the Strasbourg teaching hospital on 18/11/2009. This approval is valid for all four of the study sites in France. The protocol complies with the Helsinki Declaration and the Good Clinical Practice guidelines of the International Conference on Harmonization.

**Consent for publication**

Not applicable

**Availability of data and materials**

The datasets used and analysed during the current study are available from the corresponding author on a reasonable request.

**Competing interests**

The authors declare that they have no competing interest.
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Authors’ contributions

The project design and the different stages involved were established upstream in collaboration with the teams from the Competence and Reference Centers so as to take the specificities of each into account. BG, CD and AG participated in the conception and design of the study and its final approval, and in the drafting and revision of the manuscript. PG led the infant and family clinical assessments. CPM carried out independent, blind ratings of all available video clips of the assessments. JM & TA conducted the statistical analysis and took charge of the cleaning of the data. All the authors have read and approved the final manuscript.

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Tables

Due to technical limitations the Tables are available as downloads in the Supplemental Files.

Figures
Figure 1

Flowchart of the study
Figure 2

Evolution of ADBB scores between T0 and T1
Figure 3

Correlation matrix between instruments evaluated at T0 (left) and T1 (right) Numbers indicate Spearman correlation coefficients, ***: p<0.001, **: 0.001<p<0.01, * 0.01<p<0.05

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- Table2.pdf
- Table1.pdf
- Table3.pdf