

Relationships between parents and children with labio-palatal clefts: the importance of a delay preceding the first surgical repair



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ABSTRACT

Any attentive observer of families who have a child afflicted with a cleft lip whether or not associated with a palatal cleft would be certain to be impressed by the importance of the psychological context of their interpersonal exchanges and the risks of a breakdown in the parent-child relationship adversely affecting the child's development. The birth of a child with a cleft lip inevitably becomes a painfully stressful and traumatic event for its parents.

In the framework of the Programme Hospitalier de Recherche Clinique (PHRC) we have evaluated the psychological perceptions of parents of cleft lip and palate patients during the child's first year of life in a multi-disciplinary and multi-group prospective study. Our goal is to analyze both the child's psychic suffering and the parent-child relationship. We compared the results obtained in four hospital centers using different surgical protocols, distinguishing between the sub-groups of parents who had learned of the defect from an in utero examination and parents who did not become aware of the cleft until the child was born.

We analyze for the parents the psychic and psychological dimensions of the deformity and its correction, touching on the importance of a prenatal diagnosis, the development of a relationship with the child, self-images, and the quality of life as well focusing on the infant's distress and eventual recovery from it.

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The results of this study should make it possible for health care workers to construct new ways of attenuating, insofar as possible, the psychological impact of the deformity on parents and their children and to improve long term care for these patients.

KEYWORDS

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1 – INTRODUCTION

Cleft lips, sometimes associated with cleft palates, and isolated cleft palates are the most frequently encountered cranio-facial deformities occurring in humans, with an average incidence in the general population of one in seven hundred births³. When parents learn that their child has a serious facial defect they always suffer a major psychological shock. Often, instead of using the appellation cleft lip or cleft palate, people refer to the anomaly as a harelip, a term that

can evoke feelings of revulsion⁶, recalling for some parents of afflicted children memories of people with poor and often hyper nasal speech or with unsightly facial scars. In being presented as a “natural” anomaly, quite different from a disease, this malformation should not be seen as an accident in the life history of patients but, instead, should be accepted as a part of their essence and their destiny.

2 – PSYCHOLOGICAL IMPACT OF THE DIAGNOSIS ON PARENTS

All studies dealing with the early stages of the life of patients afflicted with labio-palatal clefts stress the importance of the quality of information that treatment team members provide to parents at their initial appointments after the diagnosis, whether it has been made *in utero* or at birth, has been announced.

When the diagnosis is pre-natal, parents are given time to accept it in stages and, thereby, to tame it pro-

gressively. But when the diagnosis is made at delivery, it is shockingly immediate.

2 –1 – Pre-natal diagnoses

Practitioners can diagnose clefts *in utero* with ultrasound examinations. Since 1980, when the first such pre-natal diagnosis was made⁴, practitioners have had constantly improving

technological means at their disposal for the detection of malformations in fetuses. Thanks to a standard number of three ultrasound exams during pregnancy which affords the opportunity to visualize the fetus before it is born, to learn its sex, the perception parents have of their unborn child is changed and so is the psychological environment of the pregnancy itself, which thus becomes a site where parents can relate to their future children and establish ties with them much earlier than was ever before possible.

By displaying their availability and their empathy and by providing pertinent information members of the treatment team can attenuate the legitimate anguish of parents of children with clefts. Many of these parents experience the birth of their child, after weeks of anxiety following the diagnosis, as a relief, especially when they learn that the defect affects only the upper lip. Parents who have benefited from the diagnosis having been made prenatally are unanimous in affirming that they are satisfied in having received the information at such an early stage and that it would have been prejudicial not to have been informed. However, it is interesting to note that when parents are asked if they are satisfied to have only learned of the malformation at the child's birth 9 to 41% of parents, according to the literature, say they would have preferred to have been informed sooner. Still most of them asserted that they were satisfied that they only learned of their child's defect at the time of its birth^{1, 9, 12, 19}. A single study has shown that all

parents would have preferred a prenatal diagnosis⁴.

2 – 2 – Post-natal diagnoses

When a diagnosis is established at birth conditions are different. The parents, especially if it is their first child, have not had time to assimilate the information and prepare for their child's deformity during the pregnancy period. Their passage into what they had conceived of as an idealized introduction to family life is transformed into a difficult and emotionally charged event. Nowadays, the time delivering mothers remain in the hospital is becoming shorter and shorter and their return to the family home, especially for mothers who didn't discover their child had a deformity until it was born, can be destabilizing¹⁴ in this period that is totally devoted to the care and feeding of the newborn. These tasks will undoubtedly seem more difficult and challenging to parents whose infant has a facial appearance that is so different from what they had imagined it was going to be and was for them difficult, even impossible to look at without apprehension. Their conception of their future family life is clearly altered by their disappointment, which can be strong enough to provoke depressive symptoms in some parents, even plunge some mothers into frank post-partum depression, social isolation, and strong feelings of culpability¹⁵. Even though very few studies have focused on what fathers of cleft palate patients suffer, it seems safe to assume that mothers must bear a preponderant share of the torment¹⁰.

3 – SURGICAL TREATMENT OF LABIO-PALATAL CLEFTS

The surgical procedures employed to correct clefts vary in accordance with the severity of the defect and the protocol subscribed to by the care giving team. At this time the ensemble of the medical community has not accepted any universal generic protocol for managing cleft palates. The clinical team uses its own protocol and its experience to make decisions about the clinical management of patients. Accordingly, the period that separates the moment when the diagnosis was made and the beginning of protocol-directed treatment, the surgical intervention, is crucial because it defines the relationship between the newborn and its parents. The deformity, especially if it takes the form of a highly visible lip defect, may adversely affect the attachment of parents to their new baby. In France, as in all of Europe, the planning for this surgical event varies widely between the various treatment centers. Some institutions favor immediate action, soon after birth in order to put the operation of the critical functions of ventilation, deglutition, and speech on a normal pathway as soon as possible and to reduce the impact of the malformation on parents and relatives. Other centers prefer to wait three to six months before operating on the newborns, believing that this added time allows growth, which is particularly active during this period, to proceed without

hindrance. It is during this stage that individualization of different muscle origins is facilitated thus improving the precision and quality of the surgical procedure. Some authors assert that these delays of various lengths might favorably condition the cognitive development of affected infants and have a positive effect of their future performance in school^{5, 18}. It has been suggested that disfigured visages are not only unattractive but that they also make it difficult for parents to interpret their child's facial expressions. So this is a major issue during the first months following birth. Murray⁸ has shown the interdependence between the child's cognitive development and the programming of the first surgical intervention. In effect, this development could be slowed down when relationships between mothers and two month-old cleft lip children can be seen to have been strained before the first operation is contemplated. No difference has been noted in parent-child relationships between control groups and cleft palate patients whose first surgery was performed at a very early date. A child's disfigurement, as we have seen, can have an adverse psychological effect on its mother and thereby harm their relationship and the length of time the child remains disfigured can, as we have also seen, explain a delay in its cognitive development.

4 – PSYCHOLOGICAL CONSEQUENCES OF THIS MALFORMATION

For many years researchers have produced studies on the psychological effects of labio-palatal clefts on both

affected children and their parents. According to a recent review of the literature⁷ 652 articles on this subject

were published between 1966 and 2003, most of which did not discern any major psychological problems. According to these papers, children with clefts did not have frank psychological, or psychiatric problems but they did suffer from behavioral disturbances, and anxiety. Moreover, both infants and their parents expressed dissatisfaction with the appearance of the infant's faces. It is difficult to interpret these signs because of the many factors that influence acceptance of this defect, including the family constellation, the severity of the cleft, the surgical protocol of the patient's treatment center, growth, and social environment.

The medico-surgical context relating to defects that affect facial appearance, an important factor in the family's inter-personal relationships and their and expression of emotions, can have a powerful effect on patients and their parents. The first interactions between parents and newborns are based on conscious and unconscious emotions that, for the most part, are expressed by touching and regarding each other's visages. When a child has a cleft, parents are brutally confronted with the infant's visage that appears broken, open, and split apart. The emotional overload of this realization can have a braking effect on their affective investment. The newborn's distorted face evokes in its family the contradictory emotional reactions of distress, horror, and blame that confront urgent wishes to protect and heal the wounded baby¹³. The malformation can interfere with recognition of an intergenerational bond and block the infant's integration into the

family network, weakening some parent-child bonds from birth onward. But some mothers have no problem establishing a fond relationship with the infant, exhibiting, perhaps, the powerful maternal urge to protect a powerless and, when its visage is damaged, an especially vulnerable child.

When members of the multi-disciplinary treatment team examine children with labio-palatal clefts, they can easily recognize the difficulties parents encounter at each stage of their children's development. They can readily visualize these problems at the time of birth and, later, at critical stages of the life cycle, like the entry into kindergarten, which will be the scene of the infant's first veritable socialization, at beginning elementary school where they will learn to read and write, and at middle and high school, a critical period when they pass through adolescence.

Treatment of labio-facial clefts requires the participation of experts from many medical and paramedical specialties and does not cease, in some cases, until patients become adults. In addition to their rupture of soft and hard tissues, labio-palatal clefts have important impacts on the functions of speech, hearing, swallowing, mastication, and ventilation and also have critical esthetic and psychological implications, affecting construction of a self-image and inter-personal relationships. Treatment team members, interacting with patients and their families, have learned to recognize the complexity of their histories and, sometimes, to assess the psychological conse-

quences of the scars left by the operations on clefts and the emotional burdens that they signify.

In attentively listening to these families over the course of the many appointments they have at treatment centers, practitioners come to understand the gravity of the trauma parents have lived through since the time they first learned of their child's deformity and the psychological context in which afflicted children participate in the parent/child relationship.

For all of these reasons, we thought it important to study what parents experience in relation to the delay between discovery of the deformity of the fetus or newborn and the first reparative surgical procedure, then to re-evaluate the experience after at least six months. It would be equally important, we believed, to seek out and analyze the emotional state of the baby with the malformation, confronted by the emotional stress its parents were undergoing.

5 – PROTOCOL OF THE STUDY

As we have previously noted, the timing of the first surgical procedure for labio-palatal cleft patients varies from treatment center to treatment center. We have selected two centers for an early intervention group and two centers for a late intervention group, Nord-Pas-de-Calais, Alsace, Ile-de France, and Lorraine.

Recruitment centers

- **For the early intervention group**

- Referral Centre for rare cranio-maxillo-facial malformations of the CHU of Lille,
- Centre Compétant of the CHU of Strasbourg.

- **For the late intervention group**

- Referral Centre for rare malformations of the face and the oral cavity of the AP-HP of Paris at the Armand-Trousseau Hospital;
- Centre Compétant of the CHU of Nancy.

Several research hypotheses

- **The principal hypothesis**

The longer the delay between birth and the first surgical operation the

more the psychological perceptions of the parents will disturb the parent-child relationship.

- **Two secondary hypotheses**

- Parents who benefited from a prenatal diagnosis of their child's labio-palatal cleft were better prepared to accept a delay of the surgical procedure;
- With time, the negative feelings of parents in the three to six month delayed surgical procedure group tended to blend in with the parents whose children had an early surgical procedure and the psychological suffering of their children also faded away.

To test this hypothesis, we assess the parents and children when the infants were 4 months old (T0) and again when they were 1 year-olds (T1).

A psychiatrist or a specially trained psychologist evaluated the psychological status and infant's capability of establishing inter-personal relationships. We then give self-administered questionnaires to parents so that the

factors capable of influencing the parent-child relationship can be assessed.

The results of the two groups will be compared, with a distinction made between the sub-groups of parents who learned the diagnosis *in utero* and those who were apprised of it at the time of birth. The judgment criteria of the first year of the child's life will deal with:

- the psychological status of the infant in relationship to the surrounding world;
- development of the parent-child bond;
- the stress endured by parents and their psychological environment;

- the development of the parent's perception of their child's deformity and their reaction to being told about it for the first time;
- the development of the family's relationship with the medical treatment team.
- Our study will deal only with children afflicted with labio-palatal clefts not those with isolated palatal clefts and no facial deformity. In order to provide valid results this study will require the participation of 150 children and their parents. It began in 2010 and should be concluded by 2012.

6 – IMPORTANCE OF INFORMATION PROVIDED TO PARENTS AND THE TYPE OF THERAPEUTIC MANAGEMENT

All studies dealing with the first stages of the life of children with labio-palatal clefts stress the importance of the information the treatment team provides parents beginning with the announcement of the diagnosis. Rey-Bellet and Hohlfeld¹² demonstrated in 2004 that a preponderant percentage of families whose child was born in an outlying maternity facility not affiliated with a hospital center complained about poorly informed and tactless staff members. Treatment teams face the problem of finding time to listen to the problems family members have and to provide them with needed information in an obstetrical or surgical department ser-

vice that, by definition, has to accommodate unpredictable emergencies in an already crowded schedule⁴. But from the very beginning parents of afflicted children need help in making an emotional investment in a child so very different from the ideal picture of their fond expectations and need to learn from the thoughtful attention and information provided by the treatment team how to become auxiliary health care providers for their babies¹¹. In their ensemble, these studies underline the prime importance of the moment when the diagnosis is announced and the caregiver-patient relationship that then begins to form.

7 – PERSPECTIVES OF THE STUDY

In its ethical aspects, we shall analyze the psychic and psychological dimensions of the situation resulting from the malformation and its correction. We shall register our database in the hope that it can serve as a reference for improving the quality of information delivered as well as the care given parents and children by the treatment team during the waiting period before an operation. We do not intend to carry out this study in a way that will intrude on medical practice or on the private lives of families. We hope, instead, that our results will serve as a platform for constructing new ways of attenuating, insofar as possible, the psychological impact of the child's deformity on its parents and to improve the treatment

of patients with labio-palatal clefts over the long term. Surgical teams dealing with this problem might also be able to utilize our data to establish, alongside other criteria, the optimal conditions for the first reparative operation. Moreover, if the conclusions derived from our research should demonstrate the benefits of offering psychological assistance to parents of children with clefts, no matter when the reparative surgical procedure takes place, a psychiatrist or psychologist member of a multi-disciplinary treatment team could propose a list of correspondents to them (recommendation of the American Cleft Palate Craniofacial Association, cited by Collett and Speltz in 2007²).

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