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Psychomotor Development of 18-Months Children With Orofacial Clefts

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Abstract

Background:

Objective – To describe the global and language development of children with cleft palate or cleft lip and palate at the age of 18 months, and to evaluate whether the type of cleft has an impact on psychomotor development.

Study Design – Prospective cohort study.

Settings – Tertiary care hospital

Patients – All children born between December 2002 and November 2009 with an orofacial cleft, operated and seen at the developmental unit (UD) of the same hospital at the age of 18 months.

Outcome Measures – Developmental quotients of the Griffiths Mental Development Scale and the French Communicative Development Inventory (IFDC) were used to assess the overall and language development of the children.

Statistics – The population characteristics were described with means for continuous variables, and frequencies for binary or categorical variables. Chi-squared and regression analysis were used to analyse the results.

Results – 69 children with clefts were examined at the age of 18 months with the IFDC and the Griffith test. The results showed that there was no significant difference in the test results of language development and global psychomotor development between the children with different types of clefts, and all were within the normal range.

Conclusion – Psychomotor development is not affected by orofacial clefts, and there is no difference between children with cleft palate or cleft lip and palate.

Keywords

Orofacial clefts, psychomotor development, language

Introduction

In Switzerland, 80 to 120 babies are born with orofacial clefts each year (Herzog, *Le Sourire aux Lèvres*, 2013). Clefts can be divided in 4 subgroups: simple cleft lips (CL),labio-maxillary clefts (LM), cleft lip and palate (CLP) and palatine clefts only (CP). The different types of clefts have different epidemiology, embryology, etiology, candidate genes, associated abnormalities and recurrence risks and can be isolated or be part of a syndrom (Wilkins-Haug, "Etiology, Prenatal Diagnosis, Obstetrical Management, and Recurrence of Orofacial Clefts", 2014).

Facial clefting may be associated with acute otitis media (AOM) and secretory chronic otitis (SCO) (Herzog, *Le Sourire aux Lèvres*, 2013), which may also impact on the child's development.

The development of children with orofacial clefts has been reported in several studies looking at different aspects: For example, Murray showed the importance of an early surgery on the relationship between the child and the parents (Murray and al., 2008), Despars the need of psychological support for the child and his family (Despars and al., 2010), and Priester and Goorhuis-Brouwer the need of speech and language support (Priester and Goorhuis-Brouwer, 2008). On his side, Swanenburg studied the relationship between associated congenital malformations and the mental and psychomotor development of children with clefts (Swanenburg and al., 2003). These studies show the importance of multidisciplinary care for children born with orofacial clefts provided by a team composed of plastic surgeons, maxillo-facial surgeons, orthodontics, speech therapists, ENT specialists and psychologists.

The aim of this study is to describe the overall and specific language development of children with cleft palate or cleft lip and palate at the age of 18 months, and to evaluate whether the type of cleft has an impact on psychomotor development. Our hypothesis is that there might be a relation between being born with a cleft and the quality of language, and that having a cleft might also affect the other developmental fields.

Methods

Population- The population consists of 69 children, operated upon by the same surgical team of a tertiary care hospital, and offered developmental follow-up. They were divided into three subgroups: cleft palate without cleft lip (CP), cleft palate with cleft lip (CLP) and a subgroup called "others" ("O"group) which included cleft lip and alveolus (CLA) or simple cleft lip (CL).

Follow-up - Developmental follow-up was offered to all the families of children operated of a cleft in our tertiary care hospital. The children were evaluated at the ages of 18 months and 5 years old. The examination at the age of 18 months consisted in a detailed history, including use of therapies, a neurological examination, developmental testing using the Griffiths Mental Development Scale, and a parental questionnaire for the assessment of language (the French Communicative Development Inventory (Inventaire Français du développement communicatif, IFDC)). IFDC, which was standardised in a population of French and French speaking Swiss children, may be used at 12, 18 and 24 months old and is a parental questionnaire which assesses expressive and receptive language of the child. It includes a list of words, which may be ticked as understood or understood and used (CD). Results are then given in percentiles.

The Griffiths Mental Development Scales, 0-2 years, is a standardized test of psychomotor development for children, which includes 5 subscales and is considered normal if it ranges between 85 and 115.
The 5 scales are

A. Locomotor
B. Personnal-social
C. Hearing-speech

D. Eye-and-Hand Coordination
E. Practical Reasoning

All the subscores can be converted with a table into a development quotient corresponding to the scale. These subscores are also added to give the final score, which is converted in the Global Development Quotient (DQ).
Source: Griffiths R. The abilities of young children: a comprehensive system of mental measurement for the first eight years of life. Association for Research in Infant and Child Development, 1984.

Data collection- Data were prospectively collected and entered in a database. The prenatal and neonatal variables were gender, gestational age, age of diagnosis and operations, comorbidities, and parental socio-economic status of the mother and father according Largo, which entails a 6 point scale for each parent, 1 being the highest (university degree or managerial work) (Largo RH, Pfister D, Molinari L, Kundu S, Lipp A, et al. Significance of prenatal, perinatal, and postnatal factors in the development of AGA preterm infants at five to seven years. *Dev Med Child Neurol*.1989; 31:440-456".). The developmental data were the results of the global development quotient (DQ) and language development quotient (LQ) of the Griffith test, and the results of IFDC at 18 months (number of words said and associated percentiles)

Statistical Analysis

The population characteristics were described with means for continuous variables, and frequencies for binary or categorical variables. We separated the population into 3 groups: CP, CLP and Other, which contain the simple cleft lips (CL) and the labio-maxillary clefts (CLA). We used the chi-squared test to compare the numerical results of the Griffith mental scales (DQ and LQ) between the different cleft groups and regression test to test the link between the quotient results and the type of clefts. The result of the chi-squared tests were considered significant if the P-value of the test was <0.05. In the regression test, the link between the two parameter analyzed was considered strong if the R-squared was between 0.4 and 0.7, and the result was consider significant if the P-value was <0.05.

Results

Population characteristics

TABLE 1 **Population Characteristics**

			CP	CLP	Other	Total
Gender	Girls		12 (44.4%)	4 (18.2%)	8 (40.0%)	24 (34.8%)
	Boys		15 (55.6%)	18 (81.8%)	12 (60.0%)	45 (65.2%)
Type	Unilateral	Left	11 (52.4%)	10 (47.6%)	21 (50.0%)	
		Right		8 (57.1%)	6 (42.9%)	14 (33.3%)
	Bilateral			3 (42.9%)	4 (57.1%)	7 (16.7%)
Largo (Mean)			3.21	3.36	2.86	3.14

There was a slight difference in the gender distribution with a majority of boys in all categories of cleft, the biggest gap being in the CLP group with 81.82% of boys.
Regarding the term of pregnancy, 90% of children were born at term (37 weeks + 1 day or more), with no difference between the types of cleft.
The following comorbidities were encountered:
In the CLP group: 1 microcephaly, 2 intra-uterine growth restriction, 2 cryptorchidism, 1 interauricular communication 1 maternal depression.
In the CP group: 4 Pierre Robin sequences, 1 left diaphragmatic hernia, 1 hamartoma of the tongue, 1 PFAPA syndrome, 1 umbilical hernia, 1 polymalformative syndrome, 1 case of dysmorphism with no identified genetic abnormality, 1 genetic strabism.
In the "Other" group: 1 interventricular communication, 1 cerebro-vascular abnormality and 1 intra-uterine growth restriction. Among these comorbidities, only 3 were related to a genetic syndrome, and there was no relation between type of cleft and type of co-morbidities.
For each child, the socio-economic status (Largo) of the parents was noted, we compared the data with the results of the DQ to see if there was any influence from the socio-economic environment on the psychomotor development according to the type of clefts, with no statistical difference between subgroups of clefts (p=0.402), but the sample size of the subgroups was too small to make any conclusion out of this analysis.
We also asked the parents when the diagnosis had been made and classified the moments of diagnosis in 3 grous: prenatal, natal and postnatal. A variance analysis of the impact of the moment of diagnosis on the DQ and LQ has been made and showed significant results for the DQ (p=0.017) but no significant results for the LQ (p=0.212). For the DQ, it appears that the best values are obtained when the diagnosis is made at birth (mean=102.68; SD=12.44).

Psychomotor development

69 children were examined at a mean age of 18 months, (SD 1.45). The neurological examination was normal in all children. As shown in Table 2, the mean and distribution of the results for the DQ was similar in the three groups.
A chi2 analysis showed no difference in the number of children with developmental delay between the 3 groups (p= 0.772, r-squared= 0.0005).

TABLE 2 Results of the DQ (Griffith scales)

DQ	n	DQ < 85 (n)	Mean	SD	Max	Min
O	20	2	101.70	12.77	123.00	71.00
CP	27	2	101.81	12.17	129.00	71.00
CLP	22	3	96.27	11.77	117.00	79.00
Total	69	7	100.01	12.31	129.00	71.00

O = Cleft lip only + cleft lip and alveolus CP = Cleft palate CLP = Cleft lip and palate

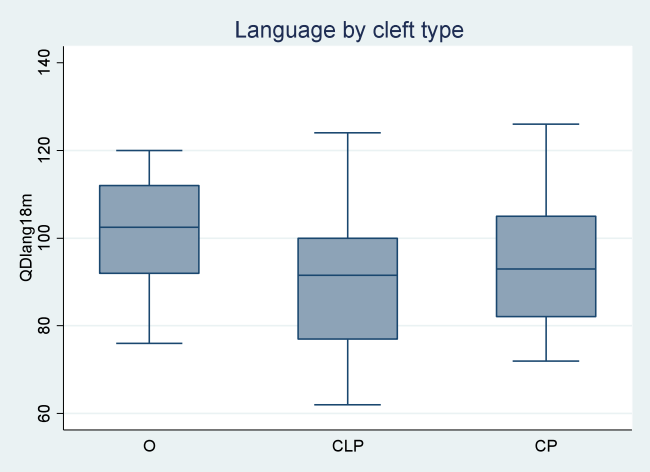
Language Development

Table 3 shows the mean (Total= 95.45, SD 14.96) and distribution of the results of the LQ.
On this analysis, the mean score of the CLP group (mean = 90.05) is lower, which is almost statistically significant (p=0.0507, adjusted r-squared=0.0587)

Table 3 **Results of LQ (Griffith scales)**

LQ	n	LQ < 85 (n)	Mean	SD	Max	Min
O	20	3	101.25	13.00	120.00	76.00
CP	27	7	95.56	14.54	126.00	72.00
CLP	22	8	90.05	15.74	124.00	62.00
Total	69	18	95.45	14.96	126.00	62.00

O = Cleft lip only + cleft lip and alveolus CP = Cleft palate CLP = Cleft lip and palate



IFDC

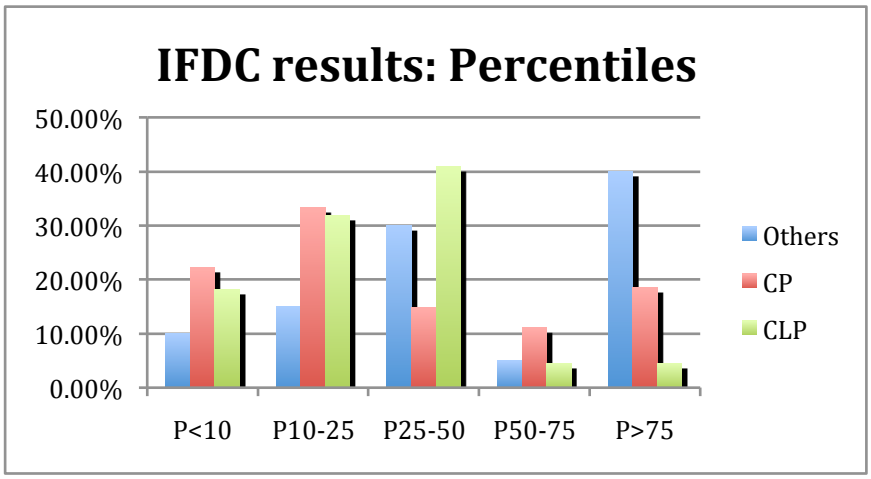
The results of the IFDC are given in number of words then converted in percentiles. As the distribution was not normal, we present the medians. The median for all the types of clefts was of 10.0 words per child. In the "Other" group the median was of 14.00 words per child, 8.00 in the CP group and 9.50 in the CLP group.

Table 4 **Results of IFDC (number of words)**

IFDC	n	Percentile	Median	SD	Max	Min
O	20	P25-50	14.00	6.79	20	0
CP	27	P25-50	8.00	6.93	20	2
CLP	22	P25-50	9.50	5.26	20	1
Total	69	P25-50	10.00	6.48	20	0

O = Cleft lip only + cleft lip and alveolus CP = Cleft palate CLP = Cleft lip and palate
n = Number of children

The following graph shows the distribution of the percentile categories depending on the type of clefts.



Others = Cleft lip only + cleft lip and alveolus CP = Cleft palate CLP = Cleft lip and palate

Discussion

The main purpose of our study was to describe language and general psychomotor development in children born with an orofacial cleft. From our practice, we hypothesized that the type of cleft might have an impact on the language and general psychomotor development of children born with a cleft.

The distribution of the different types of clefts according to the gender was in accordance with what has been previously described, CLP are most frequently observed in boys, which was the case in our database (81.82 %).

The mean results of the Griffiths Mental Development Scales are in the normal range for children with orofacial clefts. Seven children (10%) had a global developmental delay, with a DQ under 85, and 18 children (26%) a speech delay, with LQ under 85 (for these children the IFDC...). We observed that the mean DQ was slightly lower for the CLP group than for the other groups of clefts, but this difference was not statistically significant, although there was a trend. The 3 groups had a mean score in the normal range for the language score as well, with no statistical difference among them. The group of children with CLP had lower scores on both scales although not statistically significant, which could be a lack of power due to the numbers. The sample size calculation shows that 143 children per group would be necessary for the difference to be significant.

These results are in keeping with what has been previously observed in other studies. Collet and al. (2010) studied the impact of orofacial clefts on language and early reading in children from infancy through age 7. They observed no statistical difference in the language scores at age 5 and 7 between children born with a cleft and controls. Priester and Goorhuis-Brouwer (2008) chose to study the speech and language development in toddlers with or without cleft palate, and their results are in keeping with ours with no significant difference in language comprehension and production between children born with a an orofacial cleft or not. However, one study reached a different conclusion. Hentges and al. (2011) studied the cognitive development of children born with orofacial clefts with early and late surgical repair compared to children born without orofacial cleft. They observed that verbal IQ was significantly lower at age 7 in children born with orofacial cleft, irrespective of the time of

surgical repair. In our study, the age of the participants did not exceed 23 months, and the tests used to assess the language was different from this last study. Therefore these results cannot be compared to one another.

The results of the IFDC showed that the 3 groups scored in the normal range (P25-50). In the P<10 category, the most represented group was the CP group (50%). The CLP group scored most frequently under the P50 with 20 children (90% of the CLP group, 25% of all cleft groups under P50). These results are in accordance with the results of the Griffith Scales, and it appears that the more important the defect, the more consequences it has on the language tests. However, it is important to add that the IFDC is filled by the parents, not an examiner, which increases the risk of over or under-evaluation of the child capacity, and might have influenced the results.

The moment of the diagnosis might as well have an impact on the psychomotor development of the child. Our hypothesis was that if the diagnosis is antenatal, it gives more time to the parents to think about what their future child will be like and how they are going to take care of him. If the diagnosis is made at birth the parents might need time to adjust to the news and that might compromise the affective bond they need to create with their child. This bond is strongly involved in the future psychomotor development of the child, as it has been described by Murray and al. (2008) and Hentges and al. (2011). The analysis we made actually showed an impact of the moment of diagnosis on the DQ but not how we expected it. It seems that the highest DQ are found when the diagnosis is made at birth, instead of before birth. This might be explained by the fact that the clefts discovered before birth are usually cleft that involve the lip, including cleft lip and palate, and as observed before, those clefts are associated with lower DQ than the other types of clefts.

Conclusion

Our study showed that the development of children born with orofacial cleft is normal at the age of 18 months, both for global and language development. There was a non-significant difference in the results according to the type of clefts, with a lower scores if the defect was more important, but still in the normal range.

These results allow us to reassure the parents of children born with orofacial clefts on their psychomotor development thus promoting a secure parents-child relationship, which is also an important factor in the psychomotor development of a child. However, the need of multidisciplinary care for those children is not challenged. A follow-up is necessary particularly to take care of any language or psychological problems.

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