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Impact of early intravelar veloplasty at six months on mandibular growth in patients with Pierre Robin Sequence

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ABSTRACT

Pierre Robin Sequence (PRS) combines mandible microretrognathia, asynchronism of the pharynx and tongue, glossoptosis and, in some cases, cleft palate. Its principal functional consequences are respiratory and feeding problems during the neonatal period.

In this study, we focused on the impact of early closure of the cleft at six months on mandibular growth in patients with PRS.

We performed a retrospective study of 15 patients followed for PRS and undergoing surgery performed by the same senior surgeon (HB) at our cleft center between 2005 and 2012. These patients underwent early closure of the cleft (at a mean age of 5.87 months) by intravelar veloplasty, as described by Sommerlad.

Only one article with exploitable data analyzing facial and mandibular growth in a cephalometric study of children with PRS has been published. The children in this series, constituting the control group for our study, underwent veloplasty between the ages of 12 and 18 months, often accompanied by labioglossoplasty, and the cephalometric study was carried out between the ages of four and seven years. We compared this control group in which surgery was performed at 12–18 months with our series of children undergoing surgery at six months, in a cephalometric study based on teleradiographic profile measurements performed between the ages of four and seven years.

We found that early closure of the cleft soft palate yielded results identical to those for the control group in terms of mandibular growth, without the need for labioglossopexy. Finally, early intravelar veloplasty led to early functional improvement in terms of speech and phonation.

1. Introduction

The Pierre Robin Sequence (PRS) is a diagnostic trio combining glossoptosis, mandible microretrognathia, and asynchronism of the pharynx and tongue, sometimes accompanied by a cleft palate.

The timing of closure for isolated clefts palate has been well defined. Most teams close soft palate clefts towards the age of six months (Bénateau et al., 2014; Katzel et al., 2009; Martinot-Duquennoy and Capon, 2002). However, there is no consensus concerning the timing of cleft palate closure in patients with PRS. Most teams seem to close these clefts later in PRS patients than in patients with non-syndromic clefts.

Several timetables have been proposed, with two principal periods of management: early closure towards the age of six months, and later closure, between the ages of 18 and 24 months (Sommerlad, 2003). Later closure may be favored to decrease the risks associated with anesthesia, to ensure better surgical exposure and to allow the respiratory and feeding problems to stabilize.

The aim of this study was to determine whether the early closure of the cleft soft palate (at six months) in patients with PRS had beneficial effects in terms of mandible growth. Only one published article (Shen et al., 2012) has reported exploitable data for facial and mandibular growth from a cephalometric study of children with PRS. The children of this series constituted the control group for this study. They underwent cleft surgery between the ages of 12 and 18 months, often accompanied by labioglossoplasty,

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and cephalometric analyses were performed between the ages of four and seven years. We compared this control group with our series of children undergoing surgery at six months, by performing a teleradiographic study of the profiles of these children between the ages of four and seven years.

2. Materials and methods

We performed a retrospective study comparing the facial and mandibular morphologies of two groups of patients presenting PRS and a cleft soft or/and hard palate, at ages of four to seven years, after various veloplasty procedures to close the cleft.

We studied a group (group 1) of 15 patients (7 girls and 8 boys) with PRS and a cleft palate who underwent surgery in our department between 2005 and 2012. The cleft was closed by Sommerlad's intravelar veloplasty technique (Sommerlad, 2003) between the ages of five and seven months, with closure of the residual cleft palate, if necessary, between the ages of 12 and 16 months, without lateral incisions.

The control group (group 2), from a previous study on facial skeleton morphology in growing children with Pierre Robin Sequence (Shen et al., 2012), consisted of 10 patients managed at the craniofacial abnormality center of the University of California at San Francisco between 1990 and 2012. These 10 patients had PRS and presented a cleft palate. The surgical team was contacted by e-mail, and one of the authors of the article (Shen YF) provided additional information. In group 2, the cleft soft palate was closed between the ages of 12 and 18 months. Discussions with Shen revealed that most of the patients had also undergone labio-glossopexy during this operation, although this was not specified in the published article. Nevertheless, several biases remained given the lack of certain items of information: initial severity of PRS, surgical technique used, further surgery to close the residual cleft palate.

We decided to exclude patients with PRS as one of the elements of a more complex associated syndrome.

The patients of the two groups underwent teleradiographic cephalometric analyses on X rays of the entire skull, including the mandible, in profile, between the ages of four and seven years. None of the patients in the two groups studied underwent orthodontic treatment.

We ensured that the data obtained were comparable between the two groups, by performing the same type of cephalometric analysis as for the control group. We thus performed Down's analysis (Shprintzen, 1992), with the SNA and SNB angles and landmarks annotated on the cephalometric tracing (Fig. 1). This made it possible to measure the angles and lengths relating the base of the skull, the mandible and the maxilla.

Statistical analysis was performed with R software, with chi^2 tests for comparisons of qualitative data and Student's *t* tests for comparisons of quantitative data.

3. Results

In group 1, veloplasty was performed at a mean age of 5.87 months (range: 5–7 months), and cephalometry was performed at

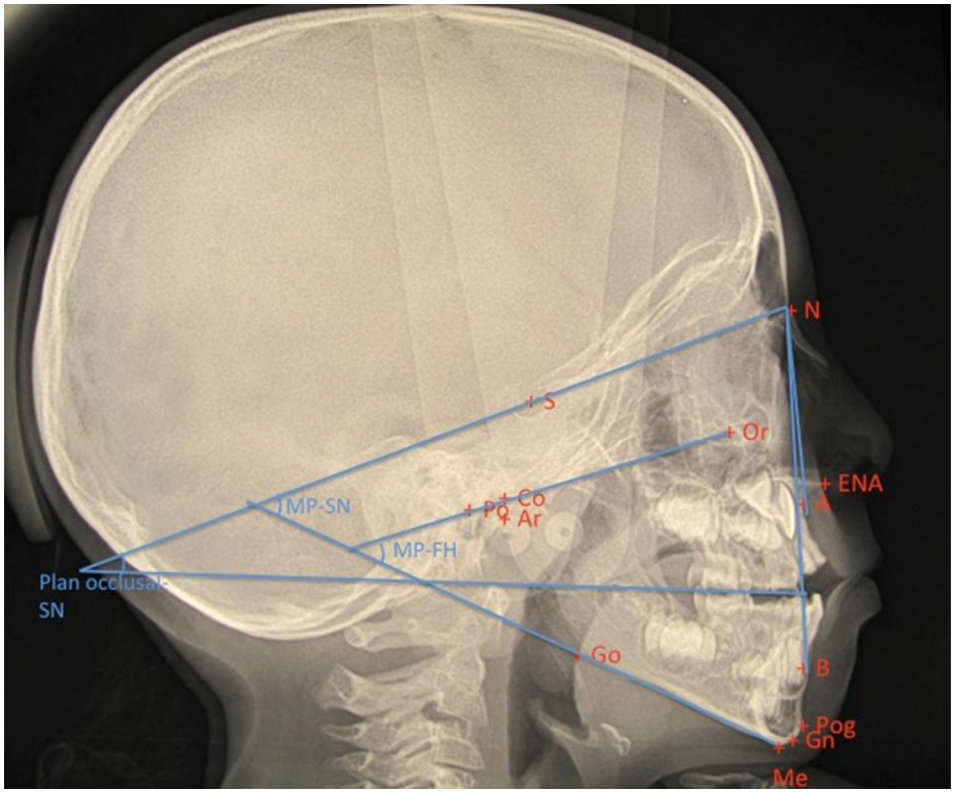

Fig. 1. Cephalometry with the key landmarks for the analysis indicated. S: Sella turcica, center of the sella turcica, N: Nasion, upper anterior point of the frontonasal suture, Co: Condylion, highest and most posterior point of the contour of the mandibular condyle, Gn: Gnathion, lower basilar point of the mandible, where the anterior curve and the basilar curve converge, Go: Gonion, the lowest point of the mandibular angle, A: A point (subspinal), most concave point on the anterior maxilla, B: B point (supramental), most concave point on the mandibular symphysis, ANS: Anterior nasal spine, Ar: Articulare, shadow between the zygomatic arch and the lower edge of the ramus, Pog: Pogonion, most anterior point of the chin, Me: Menton, lowest point of the mandibular symphysis, Or: Orbitale, lowest point on the orbital rim, Po: Porion, highest point on the external auditory meatus, The principal planes used are: The Frankfort plane (FH): plane connecting the orbitale and porion points, The occlusal plane: plane passing over the occlusal plane of the molars, The mandibular plane (MP): plane linking the menton and gonion points.