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Long-term follow-up study of patients with a unilateral complete cleft of lip, alveolus, and palate following the Utrecht treatment protocol: Dental arch relationships

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ABSTRACT

Objective: This study sought to evaluate long-term dental arch relationships in adults with a unilateral complete cleft lip and palate (UCLP) treated by the Utrecht protocol and to compare results with the centers from the Eurocleft study.

Materials and methods: Retrospective analysis of UCLP patients age 17 or older, treated by two-stage palate closure at the Wilhelmina Children's Hospital, a tertiary center for cleft surgery in Utrecht, the Netherlands. Patients were invited to the clinic for a long-term evaluation. Casts were obtained on the day of follow-up and assessed by the modified Goslon Yardstick for permanent dentition. Dental casts were scored twice by 3 different examiners.

Results: Intra-rater agreement varied from 0.743 to 0.844, the inter-rater agreement from 0.552 to 0.718. The mean Goslon Yardstick score was 3.3. Thirty-three percent of the patients had a Goslon score of 1 or 2, 45% had a score of 4 or 5.

Conclusions: The present study found unfavourable results regarding dental arch relationships after delayed hard palate closure at 3 years old. The mean Goslon Yardstick score was 3.3 (SD 1.4) and 45% of the casts were allocated to group 4 or 5 despite extensive orthodontic treatment. We observed a high number of secondary surgical interventions but no evident benefit regarding dental occlusion following the Utrecht treatment protocol, which includes a two-stage palatoplasty. Other factors than the timing of palate closure are likely of influence.

1. Introduction

The main objective of cleft lip/palate treatment is to optimize maxillofacial growth, speech development and hearing and to achieve good aesthetic outcomes. Currently no general consensus has been reached concerning the optimal timing of cleft repair (Farronato et al., 2014). It is however generally accepted that palatal surgery is one of the main factors impeding mid-facial growth in patients with orofacial clefts (Witzel et al., 1984; Semb and Shaw, 2013; Farronato et al., 2014; Johnson et al., 2014). Initially cleft palate surgery focused on immediate surgical closure of the cleft, often causing extensive dental distortions with long-term mid-face hypoplasia (Witzel et al., 1984). A trend of delaying palatal repair followed in order to diminish the amount of maxillary growth restriction (Schweckendiek, 1951; Witzel et al., 1984; Rohrich et al., 1996). Delaying palate repair however impedes adequate speech development. To circumvent this, two-stage palatoplasty was introduced in the 1950s (Schweckendiek, 1951; Bardach et al., 1984). It was hypothesized that early soft-palate repair allows for adequate speech development, while secondary delayed palate repair minimizes mid-facial growth impairment. In the following years many questioned the use of two-stage closure, mostly due to unfavourable reports regarding speech outcome (Bardach et al.,

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1984; Witzel et al., 1984; Noverraz et al., 1993; Farronato et al., 2014; Molsted et al., 2005). At present, 16.5% of cleft centers in Europe still use a two-staged approach for cleft closure, with hard palate closure between 3 and 13 years (Semb and Shaw, 2013). Studies assessing two-stage palatoplasty treatment outcomes after the completion of maxillary growth are scarce and therefore mandatory.

When evaluating the quality of cleft treatment, several outcome measures should be taken into account, including mid-facial growth. The maxillary growth is reflected in vertical, anterior-posterior and transverse dental arch relationships, making arch relationships one of the main outcomes after cleft repair. Several methods for the scoring of dental arch relationships have been put forward (Huddart and Bodenham, 1972; Mars et al., 1987; Mossey et al., 2003). Most inter-center comparisons, including the Eurocleft study, evaluated dental arch relationships using the GOSLON (Great Ormond Street, LONdon and OSlo) Yardstick (Mars et al., 1987; Molsted et al., 2005; Sitzman et al., 2014; Haque et al., 2015). More recently, an adjusted yardstick was developed to evaluate dental occlusions in patients with permanent dentition (Molsted et al., 2005; Fudalej et al., 2009; Love et al., 2012; Helena et al., 2014). This adjusted yardstick can therefore be used for scoring arch relationships in adult patients.

The aim of this present study is to evaluate long-term dental arch relationships in adults treated for an isolated complete unilateral cleft lip and palate. It will provide an insight in facial growth and dental occlusions after two-stage palatoplasty with hard palate closure at a mean age of around 3 years. Subsequently the two-stage approach will be compared to the centers involved in the Eurocleft study that used different treatment protocols.

2. Methods

2.1. Subjects

At the Wilhelmina Children's Hospital (WCH) all patients 17 years and older are invited for a long-term follow-up of their treatment as part of the standard treatment protocol. For the present study, the follow-up consultations took place between 2008 and 2014. Dental casts were obtained by the orthodontist on the day of follow-up.

This retrospective study involves non-syndromal unilateral complete cleft lip and palate patients. Medical records were searched for medical history and surgical information, including the type of surgery, technique used, age at surgery, amount of secondary surgical procedures and presence of post-operative fistulas. Both pharyngoplasties and the surgical closure of fistulas were seen as secondary, additional procedures.

Cleft repair had to comprise a two-stage palatoplasty performed by one of the surgeons of the WKZ in order to get included. Patients with incomplete medical and/or surgical data regarding the timing of soft and/or hard palate closure were excluded from analysis. Retrospectively, 78 of the invited patients met our inclusion criteria. From them 40 (51%) were included for analysis. Of the remainder, 17 did not respond to our invitation (22%), 9 were unable to attend (12%), and of 12 patients casts were not obtained during follow-up (15%). Baseline characteristics of the group with a cast were compared with the group that did not obtain a cast during follow-up (Table 2). There were no significant differences.

2.2. Surgical protocol

According to the Utrecht protocol, lip closure was performed by the Millard technique at 6 months of age (Millard, 1960). Closure of the soft palate was performed at around 9 months of age using the Perko technique (Perko, 1979). In this technique two supraperiostal flaps are dissected at the transition from hard to soft palate, without denuding bone. Nasal mucosa is sparingly mobilized and elongated by a Z-plasty. The levator palatine muscle is detached and the velar muscle complex is mobilized and reconstructed to form a transverse muscle sling. The oral mucosa is reconstructed and attached by VY-plasty. Hard palate closure was accomplished at a later age by the von Langenbeck technique (Von Langenbeck, 1861). In this bipedicle flap technique, the elevated mucoperiostal flaps remain attached to the anterior alveolar rim. Alveolar bone grafting was performed after hard palate closure at 8–12 years of age.

A pharyngoplasty was indicated when a patient had velopharyngeal insufficiency that did not improve after adequate speech treatment. The pharyngoplasty was performed according to the modified Honig technique (Mink van der Molen et al., 2009).

2.3. Orthodontic treatment

Pre-surgical orthodontic plates were only used when indicated. The indications included: feeding difficulties, abnormal tongue thrust and a clinically broad cleft. Post-surgical orthodontic obturators to cover the residual cleft were only applied in case of severe nasality impeding adequate speech development. These plates were fitted on request of the speech therapist.

Removable appliances were used for the transversal expansion of the maxilla in order to establish a normal transversal occlusion before alveolar bone grafting. A fixed appliance was used to correct the vertical plane of the central incisors. Mean age of alveolar bone grafting in the present group was 11.5 years (SD 2.5 years). Directly after alveolar bone grafting the transversal expansion was maintained for at least 6 months. After these 6 months fixed appliances were reapplied to establish the best possible occlusion. Facial masks were not used within our protocol.

Orthognathic surgery was indicated if the dental arch relationship demonstrated a negative overjet combined with either a symmetrical arch, uni- or bi-lateral cross bite or narrow arch form. This is in accordance with a Goslon Yardstick score of 4 or 5.

2.4. Dental arch relationships

In order to score the present samples the Goslon Yardstick for permanent dentition was used (Molsted et al., 2005). Models were allocated into five groups. Group 1 and 2 includes casts with a positive overjet and overbite. In group 1, a maximum of 1 tooth in cross bite was accepted. A score of 2 was given to dental casts with a good arch relationship with maximum one lateral segment in cross bite. A score of 3 was given to casts showing an edge-to-edge occlusion of the frontal region, with a uni- or bi-lateral cross bite or the inversion of one tooth of the frontal row being accepted. Casts with a negative overjet but symmetrical upper arch were given a score of 4. A uni- or bi-lateral cross bite was accepted. A score of 5 was given to models showing a negative overjet with a narrow upper arch. From a practical view, models allocated to category 4 or 5 would theoretically need orthognathic surgery for correction as orthodontic treatment alone would be insufficient. Group 3 comprised casts with fair arch relationships or malocclusions that would theoretically require complex orthodontic treatment for correction. The dental casts of patients that had orthognathic surgery ($n = 9$) prior to the day of follow-up were therefore automatically given a Goslon score of 5.

Three independent examiners scored the dental casts, namely an orthodontist (W.S.), a maxillofacial surgeon resident (G.B.) and a