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Long-term results after 40 years experience with treatment of rare facial clefts: Part 1-oblique and paramedian clefts☆

Sarah L. Versnel [a,*], Marijke E.P. van den Elzen [a], Eppo B. Wolvius [b], Charlotte S. Biesmeijer [a], J. Michiel Vaandrager [a], Jacques C. van der Meulen [a], Irene M.J. Mathijssen [a]

[a] *Department of Plastic and Reconstructive Surgery, Erasmus Medical Centre, Rotterdam, The Netherlands*
[b] *Department of Oral and Maxillofacial Surgery, Erasmus Medical Centre, Rotterdam, The Netherlands*

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Summary *Background:* Oblique and paramedian rare facial clefts impose a major reconstructive challenge and long-term assessments of the outcomes remain scarce. This study provides new details regarding surgical techniques and timing, influence of growth, and difficulties of this pathology on the long-term; a guideline for surgical treatment is given.
Methods: Twenty-nine adults with an oblique or paramedian facial cleft and surgically treated in the authors' unit between 1969 and 2009, were included. The long-term evaluation was based on series of photographs, 3D-CT's, X-rays, operation data, and was specified per facial area.
Results: The mean number of performed operations per patient was 10.6 (range: 1–26). Vertical dystopia is not caused by previous surgery, but by growth deficiencies of the maxilla. In all patients with vertical dystopia, its presence and severity were clear at the age of five, and it should ideally be treated shortly after that age. In mild cases grafting seems sufficient, but in more severe cases orbital translocation is necessary. Costochondral grafts showed the best long-term results in both orbital and nasal reconstructions. Major nose reconstruction is best delayed until adolescence. For an optimal final result in selected cases, correction of midface hypoplasia at adolescence is necessary.
Conclusion: The three-dimensional underdevelopment of the midface region plays a central role in the deformities of most patients, but is complex and difficult to correct. The provided guideline should help to minimize the number of operations and ameliorate long-term results.

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* Corresponding author. Department of Plastic and Reconstructive Surgery, Room Ee 15.91, Erasmus MC Rotterdam, Postbox 2040, 3000 CA Rotterdam, The Netherlands. Tel.: +31 10 7043291; fax: +31 10 7044685.
E-mail address: s.versnel@erasmusmc.nl (S.L. Versnel).

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Introduction

Oblique (Tessier 3,4,5) and paramedian (Tessier 1,2)[1] rare facial clefts impose a major reconstructive challenge. Often serious asymmetry exists and multiple areas of the face are affected. Due to deficient growth of all affected tissues in the cleft area, the deformities at birth can become more obvious over the years and result in clear three-dimensional underdevelopment of hard and soft tissues of the orbit, maxilla, zygoma, nose and malar region. Due to this intrinsic impaired growth or growth disturbance by surgical interventions, initial excellent treatment results may turn gradually worse. Determining the right moment and using the best technique is therefore essential.

Various techniques to address this complex pathology have been described in literature[2–7]; however, evaluation of long-term results has been scarce. Some treatment policies have become general knowledge, but evaluation of long-term results can reveal new details on which techniques give the best results or have the least relapse. It also provides additional information on the aberrant growth patterns. All previous conducted studies are limited by a small number of cases and/or a relatively short mean period of follow-up.[3–5,7–16]

This study was conducted to evaluate long-term results of surgical treatment of oblique and paramedian rare facial clefts in adults. It includes the evaluation of five cases which have been reported 25 years ago.[3] New details regarding influence of growth, techniques, timing, and difficulties imposed by this specific pathology are discussed. A guideline for surgical treatment is given.

Patients and methods

All patients with a rare facial cleft, who had surgical treatment at the Craniofacial Centre of the Erasmus Medical Centre between 1969 and 2009, were re-evaluated on initial diagnosis. All patients with an oblique or paramedian cleft were included. Patients with craniofrontonasal dysplasia, hemifacial microsomia, macrostomia, pure midline clefts (Tessier 0/14), missing data or photographs, an age less than 16 years or who were deceased, were excluded.

The series of photographs of all patients were collected. Details on performed operations were retrieved from the patient's medical chart; also when operated in other hospitals. For evaluation of the osseous structures 3D-CT's and X-rays of the patients were used when available. Since this was a retrospective study, including patients who started treatment as far as 40 years ago, only few radiological images were obtainable as quantitative parameters. Final surgical results were objectively assessed and agreed upon by three specialists, based on severity of the initial and the remaining facial deformities, using the Versnel et al. scoring list,[17] and based on the need for revisional surgery, using the Whitaker et al. classification.[18]

Results

Twenty-nine adults had an oblique (N = 22), or paramedian rare facial cleft (N = 7). Twenty patients were female. The mean age at time of follow-up was 32.1 years (SD 11.3, range 17–61), the mean follow-up was 26.4 years (SD 7.8, range 15–40). Eighteen patients had a unilateral cleft.

In general

Twenty-two patients had had operations in another hospital prior to referral. In none of the presented cases major complications were seen.

The majority of the long-term results were not as good as expected. Initially good results seemed to deteriorate over time. Patients without previous surgery in another hospital, showed better results; mainly due to better positioning of scars and superior aesthetic outcome of nose and orbital region.

At time of follow-up, nine patients were still under treatment and six restarted their treatment in consequence of their participation in our research project.

The objective severity of the total facial deformity significantly improved after treatment, as can be seen in Table 1. Looking at the specific units, also the zone of the nose and the mouth significantly improved. As for the need for additional surgery, three patients were assessed as a category I, 18 patients as category II and six patients as a category III according to the Withaker Classification.

Eyes and orbits

Soft tissue

Local flaps gave good initial results for coloboma correction. However, only a minority remained stable over time; in the majority it resulted in shortage of skin or an ectropion (Figure 1). For correction of the lower eyelid the cheek flap was superior (Figures 1–5), while the forehead flap showed tissue mismatch especially regarding thickness (Figure 2). Correction of the lateral canthus/corner of the eye was done in 19 patients; seven patients needed one or more redo's (mean: 1.7, range: 1–5). Correction of the medial canthus/corner of the eye was performed in 20 patients, of whom 16 had one or more redo's (mean: 3.2, range: 1–8). Microphtalmia was never corrected completely (Figure 3). In six patients a dacrocystorhinostomia was performed; multiple adults complained of tearing eyes.

Correction of hypertelorism

Seventeen patients had hypertelorism at birth, and in 14 patients a hypertelorism correction was performed: six medial faciotomies according to van der Meulen,[19] eight orbital box osteotomies. All the medial faciotomies were performed at an age under four. The mean reduction in interocular distance overall was 15.2 mm (range: 6–25). Six patients had an obvious residual hypertelorism after the correction due to insufficient primary correction. In two of these patients, who both had orbital box corrections, a second hypertelorism correction was performed. No relapse was seen after hypertelorism correction in the remaining eight patients, implying that growth had an insignificant influence on results of early hypertelorism corrections.