JPRAS Open 22 (2019) 9-18



Contents lists available at ScienceDirect

JPRAS Open

journal homepage: www.elsevier.com/locate/jpra

Case Report

Surgical management of the Tessier 7 cleft: A review and presentation of 5 cases

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ARTICLE INFO

Article history: Received 13 January 2019 Accepted 4 July 2019 Available online 23 July 2019

Keywords: Transverse facial cleft Macrostomia Tessier 7 Pediatric surgery

ABSTRACT

Introduction: Several variations on the surgical technique for macrostomia repair have been described in the literature. There has been controversy regarding the preferred method for commissuroplasty and skin closure for optimal functional and aesthetic results. The aim of this study is to present these techniques and the most described methods up to date.

Further, five patients operated with a combination of techniques are presented.

Material and methods: PRISMA guidelines were followed for literature review.

Five consecutive patients with unilateral macrostomia operated during a period of one and a half years at our craniofacial department were included in this study.

Results: 31 studies on macrostomia repair were obtained. The layered closure technique is widely described with several variations on closure of the inner mucosa, orbicularis muscle, commissure and skin. The inner mucosal layer is in most cases sutured with a straight line closure technique. The muscle is most often duplicated and sutured with upper branches overlapping lower branches. The skin is in most cases sutured with either a z- or a w-plasty with variations.

The five presented patients all had satisfactory functional and aesthetic results at follow-up.

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https://doi.org/10.1016/j.jpra.2019.07.004

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Conclusion: Many variations of surgical techniques for macrostomia repair have been presented in the past. We believe that each case of macrostomia needs to be assessed with a tailored surgical plan in order to create the best results. A combination of different techniques with Bütow and Botha's and Kaplan's technique as a starting point, is believed to give satisfactory functional and aesthetic results.

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Introduction

The transverse or lateral facial cleft, also called the Tessier 7 congenital cleft or macrostomia, is a rare cleft with an incidence of 1/80 000–1/300 000 live births or 0.3–1.0% of the cleft spectrum.^{1–11} It is a result of abnormal fetal development of the first and second branchial arches. It can be seen as an isolated event, in combination with other craniofacial anomalies, or as part of syndromes such as Goldenhar syndrome/Oculo-Auriculo-Vertebral Spectrum (OAVS) and Treacher-Collins syndrome.^{1–11}

The cleft is in most cases unilateral and the majority of cases does not exceed the anterior border of the masseter muscle.^{1,5,8,12-15} It involves all layers of the cheek including the skin, subcutaneous tissue, muscle and mucosa.^{1,5,8,12-16} The normal anatomy of the orbicularis oris muscle and its attachment to the risorius, zygomaticus major and depressor anguli oris muscles is altered on the cleft side.¹

Several surgical techniques for the repair of the defects seen in Tessier 7 clefts have been described. The aim of surgery is to improve feeding, speech ability and facial appearance.

Common for surgical techniques in macrostomia repair contain the following characteristics; correct markings and placement of the new commissure on the cleft side using the unaffected side as a guide, excision of excessive tissue, repair and correct placement of the orbicularis oris muscle, commissuroplasty, layered closure technique from deep to superficial layers, and skin closure using either a straight line closure, z-plasty or w-plasty.^{1-10,12-14,16-29}

In recent years, a new classification system depending on the placement and appearance of the transverse cleft was suggested by Bütow and Botha¹ (Figure 1). The authors suggest a four-layered closure technique with a superiorly based vermillion flap and a modified cutaneous z-plasty rotated superiorly for skin closure. Correct placement of the cutaneous z-plasty for normal facial expressions during smiling is emphasized (Figure 2).

In addition to the literature review, five cases of macrostomia repair using Bütow and Botha's technique, with modifications, are presented in this study.

Material and methods

The Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) – guidelines were followed as closely as possible for the systematic literature review. PubMed was searched for the following key words; "Tessier 7 cleft", "transverse facial cleft", "lateral facial cleft", "macrostomia", "macrostomia repair", "surgical treatment transverse facial cleft", "oculo-auriculo-vertebral-spectrum", "Goldenhar syndrome", "Treacher-Collins syndrome" and "hemifacial microsomia".

Studies including a more detailed description of the surgical technique and follow up on patients were included.

During a period of one and a half years at our craniofacial department, five patients with various degrees of macrostomia underwent surgery. Clinical characteristics of the five patients are presented in Table 1.



Figure 1. Butow and Botha's classification of the Tessier 7 cleft¹ (with full permission to use in this paper from Bütow and Botha, Journal of Cranio-Maxillofacial Surgery "A classification and construction of congenital lateral facial clefts").



Figure 2. Bütow and Botha's technique¹ ((with full permission to use in this paper from Bütow and Botha, Journal of Cranio-Maxillofacial Surgery "A classification and construction of congenital lateral facial clefts").

1	2
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	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5
Gender	Male	Male	Female	Male	Male
Gestation weeks	35 + 6	38 + 2	41+3	37	36 + 4
T7 cleft	Left side, 12 mm	Left side, 17 mm	Left side, 2,5 mm	Right side, 25 mm	Left side, 25 mm
Other associated anomalies	Ventricular septum defect	Goldenhar syndrome	Discrete hemifacial microsomia, ipsilateral accessory tragus, skin tags, atrophy of the parotid ductus.	Hemifacial microsomia, ipsilateral accessory ear tragus, skin tags	Hemifacial microsomia, ipsilateral accessory ear tragus, left side external ear atresia
Age at surgical procedure	7 months	14 months	6 months	8 months	5 months

 Table 1

 Clinical characteristics of the patients.

All patients were operated by the same pediatric plastic surgeons. Bütow and Botha's four-layered technique (Figure 1) with the following modifications was performed;

- (1) Mucosa excision versus preservation
- After excision of the cleft tissue, the excess mucosa has been excised when necessary. Straight line closure of the mucosa was done afterwards.
- (2) The vermillion flap The vermillion flap was raised from the upper- or lower lip, depending on where there was most excess tissue.
- (3) Muscle duplication and reconstruction of sphincter function Upper and lower muscle fibers were dissected and duplicated.
- (4) *Skin closure* The central limb of the z-plasty was placed in accordance with the nasolabial fold.

Results

Literature review

Several studies on macrostomia repair were obtained. Both systematic reviews and case reports were included. Many authors referred to the same references from the early literature and also presented arguments for their choice of surgical technique.

In Table 1 an overview of the included studies and the surgical methods described for macrostomia repair and cutaneous closure is presented. Studies may occur in more than one type of cutaneous closure category due to the use of more than one technique.

It is evident that a z-plasty or w-plasty is the most preferred cutaneous closure technique followed by a straight-line closure with or without the use of a small z-plasty at the ends. A vermillon-mucosal flap is the most often described technique for commissuroplasty. The muscle repair was in a majority of cases performed with isolation and duplication of upper and lower muscle branches. In a majority of cases, the upper muscle branches were sutured overlapping the lower branches. The inner mucosal layer is most often sutured with a straight line closure (Table 2).

Studies originated from different parts of the world and included mostly unilateral macrostomia cases.

Cases

Five patients underwent surgery; four males and one female. All patients were operated with Bütow and Botha's modified technique as described at an age of 6–14 months.

The z-plasty for skin closure was obtained with satisfactory aesthetic results in all cases. No scar hypertrophy, contracture or migration of the commissure was observed. No postoperative

Table 2

Overview of the studies included in the literature review and the surgical techniques presented.

Commissural closure	Cutaneous closure				
	Z-plasty	W-plasty	Straight line		
Linear	Longacre, ¹² Boo-Chai, ¹⁷ Mansfield ²⁶				
Vermillon-mucosal flap(s)					
Rectangular					
Inferiorly based	Aketa ²⁴	Dhingra ³	Weinstein, ²⁷ Rogers ⁸		
Superiorly based	Butow, ¹ Kaplan, ¹⁴ Li ¹¹	Eguchi ²⁰	-0		
Inferiorly and superiorly based	Makhija, ⁹ Verheyden ¹⁸	U			
Triangular	5. 5				
Inferiorly based	Jaworski ²⁵				
Inferiorly and superiorly based	Chen ³⁶	Bauer ³⁵			
Inferiorly and superiorly based	Kajikawa, ¹⁰ Kobraei ⁵	Habal ²²	Kajikawa, ¹⁰ Popescu ¹⁶		
Cutaneous flap					
Triangular			Kawai ⁴		
Inferiorly based	Yu, ⁶ Ono, ²⁸ Yoshimura ¹⁹		Yoshimura ¹⁹		
Myomucosal flap and cutaneous flap, either inferiorly or superiorly based	Torkut ²⁹		Franco ⁷		
Advancement flap – transposition of cleft	Fukuda ²¹	Fukuda ²¹			
(vermillon-mucosal-cutaneous flap)					
Full-thickness vermillon-mucosal-cutaneous flap, triangular (Estlander flap)	May ¹³				
Z-plasty	Kuriyama ²				



Picture 1. Patient 1 – preoperative and 3 months postoperative image.

complications were observed. The patients were evaluated three months postoperatively. The appearance of the scar and the functional results were evaluated by the surgeons and the parents.

Preoperative and postoperative images are shown in Pictures 1-5.

Discussion

Tessier 7 clefts are rare with a reported incidence of 1/80 000–1/300 000 live births. The most often described anomalies seen in combination with the Tessier 7 cleft are OAVS encompassing the Goldenhar syndrome and hemifacial microsomia, and Treacher Collins syndrome.^{1,5,11,15,30,31} OAVS shows a preponderance in males with a ratio of approximately 3:2.^{30,31} The most common craniofacial anomalies associated with OAVS are ear deformities, epibulbar dermoids, orofacial clefts and hemifa-



Picture 2. Patient 2 - preoperative and 3 months postoperative image.



Picture 3. Patient 3 – preoperative and 3 months postoperative image.



Picture 4. Patient 4 – preoperative and 3 months postoperative image.



Picture 5. Patient 5 - preoperative and 10 months postoperative image.

cial microsomia with various degrees of underdevelopment of the maxilla and mandibula.^{30–32} Other than craniofacial anomalies, OAVS patients often present vertebral deformities and congenital heart defects.^{30,31}

Treacher–Collins syndrome is an autosomal dominant congenital disorder. The incidence is approximately 1 in 50 000 live births.³³The patients present craniofacial anomalies in various degrees.^{33,34} The most common described craniofacial anomalies are defects in the periorbital region with a typical downward displacement of the lower eyelid, hypoplasia of the mandibula and zygoma, dental anomalies and ear deformities often accompanied with hearing disabilities.^{33,34} Macrostomia is an occasional finding in Treacher–Collins patients.^{33,34}

The macrostomia seen in patients with congenital disorders should be assessed as early as possible to facilitate normal development in speech, eating and facial appearance.^{15,32} Delayed repair for a more accurate placement of the new commissure has been suggested in the early literature in patients where mandibular and zygomatic repair is necessary.¹²

Amongst the first surgical techniques for macrostomia repair, an Estlander flap with a full thickness vermillion lined flap was described.¹³ A layered closure technique was used, however, the importance of the correct adaptation and placement of the orbicularis oris muscle was not described until later.¹⁷ The correct placement of the new commissure as well as skin closure are two important features of surgery which have been more widely described recently.

Boo-Chai described a change in vermillion color and thickness and in the white line in the cleft.¹⁷ The importance of correct placement of the orbicularis oris muscle as close to the new commissure as possible was also first described by Boo-Chai in order to avoid a "goldfish mouth" appearance.¹⁷ The relation to the eyes and pupils, which correlates to the commissure, was also described in the early literature.^{14,17,26}

The four-layered technique with several variations was subsequently introduced.^{14,24–27} In 1981, Kaplan introduced a modified technique for preoperative markings and surgical procedures. A rectangular vermillion-mucosal flap based superiorly was described. The overlapping of upper muscle bundles on lower muscle bundles was described to facilitate the natural "overhang" appearance of the upper lip in relation to the lower lip. This is however not described as mandatory. Kaplan advocates a z-plasty for skin closure to avoid lateral and downward displacement of the scar. The central limb of the z-plasty should be placed in accordance with the nasolabial fold. However, soft tissue- and bone deficiency are also mentioned as reasons for lateral and downward displacement of the new commissure.¹⁴

Since then, several papers have been published with a variety of techniques for a layered closure of macrostomia. $^{4,6-8,10,11,18-22,28,35}$

Functional and aesthetic outcome including avoiding lateral and downward displacement of the scar have been discussed in the literature.^{1,4–6,8,14,18–22,35} Straight line skin closure is believed to result in contracture, lateral displacement of the commissure and poor aesthetic results^{1,4–6,14,18–21,24,35}.

However, actual studies have not proven this. Additionally, the lateral displacement caused by a straight line skin closure is in some studies believed to be prohibited by a constant medially directed dynamic "counterforce" by the sphincter function of the orbicularis oris muscle, once repaired.^{8,13} Interestingly, there are only very few cases worldwide where poor functional and aesthetic outcomes with a straight line closure are described.^{4,8,19} Two more recent studies have shown satisfactory functional and aesthetic results with a combination of vermillon-mucosal flaps for commissure closure and a straight line skin closure.^{8,10} However, a vast majority of the studies in the literature include skin closure with either a z-plasty or a w-plasty. Some advocate a w-plasty for skin closure because it is believed to create a less conspicuous scar than a z-plasty.^{22,35} Furthermore, some authors argue that the size of the z-plasty is believed to affect the final aesthetic result.⁸ Larger z-plasties (>10 mm) is believed to result in more conspicuous scars than smaller z-plasties (< 6 mm).⁸ A straight line closure with smaller z-plasties at the ends of the scar to avoid dog ear formation have also been presented.^{4,19}

During later years, the optimal placement of the limbs of the z-plasty have been discussed.^{5,6,8,10} Whether the central limb of the z-plasty should be placed in the nasolabial fold for a better aesthetic result and with less tension is still debated. Some authors argue, similar to Kaplan, that migration of the commissure is not necessarily due to poor skin closure, but poorly executed muscular- and vermillion closure.^{2,4,8,14}

As for the commissuroplasty as well, there has also been many variations in the literature.^{1,4,12–14,20–22,26–29,35} Amongst these, a superiorly based vermillon-mucosal flap with a turnover mucosal flap for intraoral lining prior to muscle overlapping and z-plasty for skin closure has been suggested.¹⁴ Several variations have been presented since. Both rectangular and triangular vermillonmucosal flaps raised either superiorly or inferiorly are the most described in the literature.^{8,10,20,35}

The most well described intraoral mucosa closure is either an intraoral z-plasty or a straight line closure.

Our described technique and modifications are based on that each cleft is unique and should be assessed according to the anatomy of the cleft. Therefore, we have not set a fixed method for each step of the surgical procedure. Similar to Butow and Botha's technique, each cleft is assessed based on the unique appearance i.e., width, direction and length of the cleft, which should be taken under consideration during surgical planning.

The presented technique is preferred as it gives a pleasant and consistent result. As the anomaly is rare, it is important to have the surgical details specified in order to avoid unsatisfactory cosmetic and functional results. It is our opinion that Z-plasties give at better result both cosmetically and functionally in the long-term compared to the straight line technique. Also, the Z-plasties should have a certain length, i.e., about 1 cm, as smaller Z-plasties tend to look and have the function of a straight line.

The vermilion tissue is transposed from the upper lip, around the commisure and to the lower lip to avoid a suture line directly in the commisure, as it is our experience that this may cause delayed healing or chronic ulceration. A disadvantage of this could theoretically be that the vermillion does not narrow towards the commisure as it would in natural circumstances, but according to our postoperative evaluations this detail is not clinically obvious and hence is not considered important.

As to muscle duplication, we have similar to Kaplan's technique performed an overlapping of the upper and lower bundles of the orbicularis oris muscle.¹⁴ The reconstruction of the modioulus with suturing of the orbicularis oris, zygomaticus major, risorius and depressor anguli oris muscles as described by Buthow and Botha,¹ has not been performed in our cases.

A limitation in this study is the short follow-up period. Further follow-up by our team is planned and the patients are followed by a multidisciplinary team to optimize speech abilities and monitor facial growth. If required, further surgery will be planned in order to optimize maxillary and mandibular growth.

Conclusion

Macrostomia repair is well described in the literature. Many variations of surgical techniques have been presented in the past. There is still controversy as to which skin closure has the best functional and aesthetic result. The commissuroplasty has also been described with many different techniques. We believe that each case of macrostomia needs to be assessed with a tailored surgical plan in order to create the best results. A combination of different techniques with Bütow and Botha's and Kaplan's technique as a starting point, is believed to give satisfactory functional and aesthetic results.

Funding

None.

Declaration of Competing Interest

None.

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