Isolated bilateral macrostomia: literature review and case report

M. Tarle[1,2], A. Tarle[2,3], D. Macan[1,2], H. Knežević Krajina[4], P. Knežević[1,2]

[1]Department of Oral and Maxillofacial Surgery, Dubrava University Hospital, Zagreb, Croatia
[2]University of Zagreb, School of Dental Medicine, Zagreb, Croatia
[3]Apolonia Dental Clinic, Zagreb, Croatia
[4]University of Zagreb, School of Medicine, Zagreb, Croatia

E-mail: tarleantonia@gmail.com

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Abstract

Aim Macrostomia, or lateral cleft lip, which is known as Tessier cleft type 7, is one of the rarest facial anomalies. The purpose of this review is to describe the main characteristics, epidemiology, aetiology and treatment of this anomaly.

Methods We present an overview of surgical techniques as well as a review of all 36 cases of bilateral asyndromic macrostomia reported to this da in the literature. Furthermore, we report the case of a 4-month male infant with bilateral transverse cleft lip and analyse the treatment decision and the procedure itself.

Results Macrostomia may be found as a part of syndromes like oto-mandibular dysostosis, hemifacial microsomia, Treacher-Collins or Goldenhar syndrome, or in conjunction with additional facial symptoms. However, it is uncommon to find macrostomia as an isolated asyndromic entity. There are many surgical techniques proposed for the reconstruction of macrostomia, yet there is no consensus on the gold standard.

Conclusion Early diagnosis and surgical intervention are crucial in treating children with these malformations. Adequate timely reconstruction plays a main role in both physical and psychological rehabilitation.

KEYWORDS Macrostomia, Tessier number 7, Isolated bilateral macrostomia.

Introduction

Facial clefts are the most common facial anomalies and cover a broad variety of soft and bony tissue defects [Habel et al., 1996; Rullo et al., 2014]. Lateral, or transverse, cleft (macrostomia) is one of the rarest types and according to Tessiers classification is referred to as the Tessier type 7 [Tessier, 1976]. The incidence of this atypical malformation is 1 in 80,000 to 1 in 300,000 of all live births and only 0.3–1% of all facial clefts, including cleft lip [Raveendran et al., 2018]. The defect can occur unilaterally, which is significantly more common, while bilateral macrostomia can be seen in only 10–20% of the cases and is usually symmetrical [Buonocore et al., 2014]. Macrostomia is more common in male babies and in the unilateral form. This anomaly is often combined with syndromes, however, the asyndromic type, also referred to as isolated, is very uncommon. It is often part of syndromes like oto-mandibular dysostosis, hemifacial microsomia, Treacher-Collins or Goldenhar syndrome, or in conjunction with additional congenital facial deformations such as preauricular tags, zygomatic arch deficiency, deformities in different mandibular parts such as ramus, condyle, or coronoid process, or ear anomalies (deformities of the external, middle or inner parts, eustachian tube absence), and other facial clefts such as Tessier 2-5 [David et al., 1987]. Bilateral macrostomia, despite being rare, occurs as asyndromic more often than the unilateral cleft [Gleizal et al., 2007]. The cause of congenital macrostomias remains unclear but genetic factors, inadequate blood supply of the brachial arch, uncontrolled apoptosis, damage to the stapedial artery, presence of the amniotic fluid bands and amniotic tenting are often mentioned when trying to explain the aetiopathogenesis [Buonocore et al., 2014]. Prenatal ultrasound detection of facial clefts is often unsuccessful in noticing macrostomia [Cavaco-Gomes et al., 2017], as it was in our case.

The purpose of this work was to systematically show the classifications offered in literature and present and compare different surgical treatment methods, as well as to show current scientific theories regarding the causes and present an up to date review of the published cases. The case we present is a 4-month-old Caucasian male infant who was referred to our Department for correction of symmetrical bilateral transverse cleft lip.

Literature review

A literature review was conducted by searching the PubMed, Scopus and Scholar database using the keywords "macrostomia", "bilateral macrostomia", "isolated bilateral macrostomia", "bilateral transverse facial cleft" and only 37 cases have been reported (Table 1). Cases included in the review were only bilateral isolated macrostomia without bone involvement. We notice that the majority of cases (7:1) were not born of a first pregnancy which may be useful for understanding the aetiology. There were slightly more females than males (19:16).

Macrostomia is a rare malformation, therefore rarely described in literature and sometimes very confusing to classify. In comparison to typical facial clefts (cleft lip and cleft palate) all the remaining clefts are part of the atypical group and take up only 3% of all facial clefts [Cavaco-Gomes et al., 2017]. Macrostomia is most commonly a synonym for transverse facial clefts also referred to as lateral or commissural facial cleft. This describes a variety of defects that extend along an imaginary line connecting the oral commissure and the tragus, and result in an enlargement of the mouth [Eppley et al., 2005]. The malformation may present unilaterally or bilaterally, as an isolated entity or as a part of syndromes and combined with other congenital facial

No.	Author	Year	No. of cases	Sex	Ethnicity	Skin closure	Born from first pregnancy
1.	Powell and Jenkins	1968	1	F	Caucasian	no data	no data
2.	Hawkins et al.	1973	1	F	Caucasian	Z-plasty	+
3.	Talukder	1980	1	M	Indian	no data	no data
4.	Bauer and Wilkes	1982	1	F	Caucasian	W-plasty	no data
5.	Habal and Scheuerle	1983	1	F	Caucasian	W-plasty	no data
6.	Fukuda and Tadeka	1985	1	M	Asian	Z-plasty and W plasty	no data
7.	Aznard	1989	1	M	Caucasian	no data	no data
8.	Beziat	2007	2	1F 1M	Caucasian	straight line	no data
9.	Vazquez	2007	3	1F 2M	African, Caucasian	Z-plasty	no data
10.	Nathani	2008	1	F	Indian	Z-plasty	-
11.	Fadeyibi et al.	2010	4	3F 1M	African	Z-plasty	no data
12.	Ahmed et al.	2010	1	M	Indian	W-plasty	-
13.	Sowande et al.	2011	2	2F	African	Z-plasty	-
14.	Khaleghnejad-Tabari et al.	2012	1	F	Caucasian	straight line	-
15.	Mohan et al.	2013	1	F	Indian	no data	no data
16.	Oghale	2013	1	F	African	Z-plasty	-
17.	Buonocore et al.	2014	3	1F 2M	Caucasian	no data	no data
18.	Narendra	2014	1	F	Indian	no data	no data
19.	Pradhan et al.	2018	6	2F 4M	Asian	Z-plasty or straight line	no data
20.	Tse et al.	2018	2	no data	Caucasian	straight line	no data
21.	Wong et al.	2018	1	M	Caucasian	Z-plasty	-
22.	Knežević et al.	2020	1	M	Caucasian	straight line	-
TOTAL			37	19F 16M			

TABLE 1 Clinical details of all patients with isolated bilateral macrostomia in reported literature.

deformations [Lezama-Reus et al., 2007]. The clinical presentation of the deformity may vary from an unnoticeable widening of the oral aperture to a full thickness defect comprising the skin, muscle and bony tissue [Lewin, 1950].

Classification

The first anatomical classification of facial clefts was made in 1976 by Tessier, who divided the malformations into 15 types based on the direction of the cleft relative to the orbit [Tessier, 1976]. According to this classification, the defects that have a trajectory placed laterally to the infraorbital foramen are considered lateral clefts and are assigned the numbers 5–9 [Bradley and Kawamoto, 2013]. The Tessier type 7 cleft involves soft tissue and bony component, otherwise referred to as temporo-zygomatic cleft centered on the zygomaticotemporal suture [Butow and Botha, 2010]. Considering that this classification differentiates types based only on topography, other systematisations emerged, such as the one by Van der Meulen in 1983 based on embryogenesis and aetiopathogenesis, which describes the lateral type 7 cleft as a maxillo-mandibular dysplasia, a failure of fusion of the maxillary and the mandibular process. According to this systematisation, lateral cleft lip implies soft tissue involvement with or without preauricular fistulas or appendages. Bony involvement affects the pterygomaxillary junction, or includes hypoplasia of the alveolar process, maxilla, zygomatic bone, mandibular processes or the sphenoid bone [Butow and Botha, 2010; David et al., 1989]. Subsequently, other subclassifications were introduced to complement the latter and aid to optimal protocol management. David et al. [1987] recommended an addition to the classification:

- Tessier 7a: Maxillary cleft;
- Tessier 7b: Maxillary duplication. According to the maxillary involvement, a bony malformation presenting either as hypoplasia or overlapping of maxillary arches (duplication) and additional teeth [Woods et al., 2008].

Variations in appearance encouraged Butow and Botha to suggest that Tessier type 7 should be divided into 4 subtypes:

- 7.1a for the superiorly rotated cleft without bony involvement and 7.1b including bony malformations;
- 7.2 denoting middle-positioned clefts with subdivisions for bony involvement (7.2a with and 7.2b without);
- 7.3 for the inferiorly rotated; and
- 7.4 for the agenetic lateral facial cleft with same subdivisions for bony involvement (however, such cases are yet to be reported).

The Tessier 7.1 includes separation of the zygomatic major and risorius muscle, 7.2 clefting of the risorius muscle, 7.3 separation of the risorius and depressor anguli oris muscles, while 7.4 involves agenesis, or partial agenesis of the risorius muscle, with or without partial agenesis of the orbicularis oris [Butow and Botha, 2010]. Finally, Gleizal et al. [2006] proposed a classification into 4 types with therapeutic implications:

- type I or minor unilateral macrostomia: the cleft terminating medial to the masseter border;
- type II or major unilateral macrostomia: the cleft extends to the tonsillar pillars (IIa) or into the tragus (IIb);
- type III or bilateral minor macrostomia (same as unilateral);
- type IV - bilateral major macrostomia with sagittal cleft extension (IVa) or transverse cleft extension (type IVb) [Gleizal et al., 2007; Mohan et al., 2013].

Aepidemiology

Tessier type 7 cleft is one of the rarest types of anomalies, but at the same time the most common atypical cleft. It occurs in 1 in 80,000 to 1 in 300,000 of all live births and only 0.3–1% of all facial clefts, including cleft lip [Raveendran et al., 2018]. Bilateral macrostomia can be seen in only 10–20% of the cases and shows male predilection, while the unilateral form occurs 6 times more often and slightly more frequently on the left side.

Bilateral macrostomia is usually symmetrical [Buonocore et al., 2014; Kuriyama et al., 2008]. This defect is often combined with syndromes, most commonly Treacher-Collins, Goldenhar, oto-mandibular dysostosis and hemifacial microsomia. The asyndromic type, also referred to as isolated, is very uncommon. Although rare, the bilateral macrostomia, occurs as asyndromic more often than the unilateral type [David et al., 1987].

Aetiology

The aetiology of macrostomia is unknown to this day. There are many theories that tried to clarify the aetiopathogenesis. It is assumed that both environmental and genetic factors play an important role. The first theory proposed describes the failure of fusion of the maxillary and mandibular processes resulting in a defect running from the oral commissure to the tragus [Van der Meulen, 1985]. This fusion normally occurs at week 6 of embryonic development but the failure to fuse is not yet explained [Buonocore et al., 2014; Van der Meulen, 1985]. No specific growth factors have been linked to the fusion of the maxillary and mandibular processes, however, FGF8, produced by the epithelium, seems to play an important role in the proximal portion of the first brachial arch providing survival, mitogenic, antidifferential and patterning signals to the mesenchyme of the proximal arch [Moon and Capecchi, 2000; Beerman et al., 2006]. Reduced expression of this fibroblast growth factor in the first arch ectoderm results in increased apoptosis and decreased proliferation of first arch neural crest mesenchyme [Lewandoski et al., 2000]. Fgf8 gene inactivation in chick embryos results in craniofacial abnormalities such as first branchial arch hypoplasia which is enough to consider this as an important factor in the aetiopathogenesis [Trumpp et al., 1999].

Another important factor is intrauterine trauma, such as amniotic band syndrome and amniotic tenting in association with intrauterine synechiae [Dashe et al., 2002]. Entrapment of the fetal structures by mesodermal bands originated from chorionic side may be caused by early rupture of the amnion and can result in malformations. There is evidence that lateral clefts may occur at stages post-organogenesis rather than earlier due to amniotic bands [Presti et al., 2004]. *In utero* animal model tried to demonstrate a relationship between atypical clefts and restricting forces imitating the bands by attaching nylon sutures to the zygomatic arch or fronto-orbital rim in fetal lambs. The bands caused lateral cleft macrostomia in all the animals in less than 140 days [Stelnicki et al., 1997]. The timing of fetal entrapment may also be of significance. Formation of bands before week 6 of gestation will lodge between the facial processes and cause clefts that follow the embryonic fusion lines. The same mechanism occurring after 6 weeks when the face is fully formed causes oblique clefts and nerve palsies by disrupting the fetal tissues [Tharanon et al., 1998]. Intrauterine trauma, curettage procedures most commonly performed after abortions, caesarean section, IUD and uterine surgery are the leading causes of amniotic tenting and synechiae [Presti et al., 2004]. The proposed vascular aetiology theory due to the presence of hematoma in the territory of the stapedial artery preventing the fusion of the processes is considered less likely in bilateral macrostomia because both arteries would need to be affected [Gleizal et al., 2007]. Early prenatal detection of asyndromic transverse facial cleft proved to be very difficult as it has been described in literature in only a few cases. A detailed morphological ultrasound is crucial in detecting these malformations and 3D-US is a much superior method to the standard 2D-US in understanding the anatomy of the defect [Cavaco-Gomes et al., 2017].

Surgical treatment

Asyndromic bilateral macrostomia presents a significant challenge for the surgeon because even very experienced surgeons will be confronted with no more than a few cases during their career. The satisfactory result of the surgical procedure cannot be achieved without completely restoring function of orbicularis oris muscle but at the same time achieving high aesthetic goals—natural looking commissure and a minimally visible scar. Besides function and aesthetics, it is imperative to keep in mind the psychological effects of the cleft malformation on the child but also on the family, therefore the earliest possible surgical intervention is recommended [Butow and Botha, 2010]. The functional issues caused by dysfunction of the orbicularis muscle include sialorrhoea, compromised speech and chewing ability, and difficulty sucking [Gunturu et al., 2014]. Although the literature offers different opinions on the best surgical solutions for resolving these problems, the goals are always: 1) restoration of the oral sphincter and other facial muscles; 2) creating a functional and natural looking oral commissure; 3) achieving symmetrical lips without a visible discontinuity of the vermilion border; 4) closure of the skin defect with achieving minimal scar; 5) multi-layer closure of the defect [Raveendran et al., 2018; Butow and Botha, 2010; Gunturu et al., 2014; Yencha, 2001].

The first surgical attempt at treating macrostomia described in literature was by May in 1962 who used the Eastlander flap. The same year a trilangular excision followed by a trilaminar linear closure was described as a technique for treating macrostomia [Kobraei et al., 2016]. The method was faulty mainly because placing the scar at the commissure resulted in contractures, fissuring and unsatisfactory appearance. The uneven vermilion border caused soakage of the wound which led to erosions, cheilitis and tearing while opening the mouth. Successful reconstruction of the commissure without these complications became the focus of developers and therefore various flaps designs were introduced [Raveendran et al., 2018; Kobraei et al., 2016; Rogers and Mulliken, 2007]. Boo-Chai began the advancement by identifying the vermilion border in 1969 recognising the difference in colour and a mark on the border of the vermilion at the location of the neocommissure. He described the importance of identifying orbicularis oris borders and positioning them as close as possible to the neocommissure [Boo-Chai, 1969]. The first commissural flap was described by Kaplan in 1981 when he transposed a square vermilion flap from the upper lip and connected it to the lower lip leaving the suture not in the commissure, but on the lower lip, successfully avoiding the already described complications [Kaplan, 1981].

While reviewing the literature, it becomes apparent that there has not been a consensus on the best surgical method for skin closure [Kobraei et al., 2016]. While simple linear closure appears to leave the most inconspicuous scar and has pleasing aesthetic results, it is criticised for having increased risk of inferior and lateral commissure migration [Kobraei et al., 2016; Yoshimura et al., 1992]. As an alternative, The Z-plasty and W-plasty along with combination techniques emerged and are still claimed to be superior to the straight-line technique by many authors [Kobraei et al., 2016; Kaplan, 1981]. The Z-plasty skin closure technique is supposed to stabilise the commissure, but lengthens the scar itself, sometimes resulting in a displeasing appearance. This was reduced with the W-plasty skin closure which was modified in 2001 and described as a lazy W-plasty used for skin closure after a vermilion square flap by Eguchi et al. [2001]. Their square flap was raised off the lower lip so the scar would rest on the upper lip as they believed that lower lip scars

become more conspicuous over time due to tension. Ono et al. reported a technique using two triangular skin flaps combined with suturing the orbicularis to the risorius or buccinator to reinforce the muscle [Ono and Tateshita, 2000]. A slightly different approach to muscle reconstruction was offered by Franco et al. in 2007, when he described a myomucosal flap technique that does not separate the muscle from the mucosa but repairs the muscle using end-to-end connection combined with the vermilion flap and straight-line closure ensuring ideal lip thickness and muscle function [Franco et al., 2007; Tse et al., 2018; Ueno et al., 2021]. However, many other authors believe the overlapping muscle repair to be a preferable option for achieving more aesthetic goals and avoiding the unwanted "goldfish mouth" [Kaplan, 1981; Eguchi et al., 2001].

Based on the authors significant clinical experience in cleft surgery (especially unilateral transverse clefts) in a high-volume cleft center at the Dubrava University Hospital and on cases shown in literature, we decided to use the square vermilion flap combined with the end-to-end muscle repair and straight-line skin closure. Although mentioned to carry more risk of scar contracture, we believe that, if executed meticulously, this technique may bring aesthetically far more pleasing results without compromising the healthy areas and leave the possibility of easy revision. A great variety of surgical methods and techniques shows that there is no gold standard to treat this rare deformity and satisfactory results can be achieved with various techniques.

Case report

A healthy male newborn was presented to our clinic 5 days after birth with congenital bilateral macrostomia. The baby was born full term at 40 weeks of gestation through vaginal spontaneous delivery, to a 38-year-old mother. The baby was 52 cm long and weighing 3,660 g, Apgar score 10/10. The obstetric history of the mother (gravida 3 para 2) shows one previous delivery of a healthy female child and one miscarriage in the first trimester. She had a history of gestational diabetes and uterine septum in the first pregnancy. Prenatal diagnostics included regular ultrasounds with a finding of a uterine septum, the Harmony test which stated a low risk of chromosomal abnormalities and a combined screening test that indicated a borderline result (1:250). During the pregnancy, the mother took dydrogesteronum and folic acid. Parents were not relatives, with no family history of craniofacial anomalies.

The extraoral clinical exam revealed a symmetrical bilateral transverse facial cleft extending 12 mm laterally from the vermilion border (Fig. 1). The malformation was middle positioned, clearly defined from the vermilion border, lined with mucosa buccally and skin externally including diastasis of orbicularis oris muscle without reaching the anterior border of the masseter. The radiological examination confirmed there were no skeletal deformities and the intraoral examination showed a symmetrical normally developed alveolar ridge, palate, and soft tissue. The function was not impaired and sucking and breastfeeding abilities were not disturbed. The chief complaint of the mother was the unsightly appearance as well as saliva flowing outside of the mouth. Further paediatric diagnostics included an ultrasound of the abdomen, the hips and the brain, an ophthalmological examination, and a genetic screening which all showed no pathological findings. The final diagnosis was Tessier type 7 isolated bilateral cleft.

FIG. 1 Pre-operative appearance of bilateral macrostomia symmetrically extending 12 mm laterally from the vermilion border.

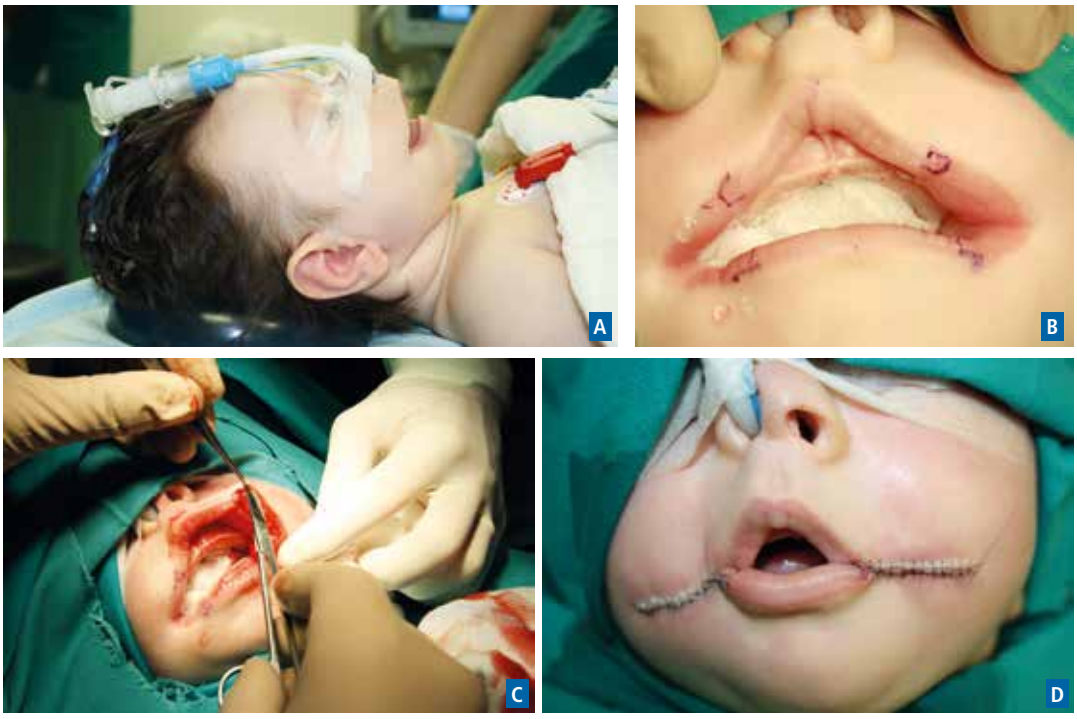

FIG. 2 Surgical procedure.
A: Endonasal intubation.
B: Planning of neocommissural position and square flap design.
C: Skin is carefully dissected from the muscle and buccal mucosa. The layers were then connected separately using single stiches with Vicryl 4-0 suture.
D: Straight-line skin closure using Prolene 6-0 suture.

FIG. 3 Six-month postoperative appearance.

The treatment decision was made after an extensive literature research. The vermilion border was visible and easily distinguished from the defect, extending equidistant from the midline, but still marked preoperatively. The surgery was performed on the three-month-old patient in general anaesthesia with nasotracheal intubation to ensure an undisturbed access to the lips and to be able to precisely mark the vermilion border. A straight-line cutaneous closure was used to close the cleft combined with a square vermilion flap surgical technique to ensure a natural commissure. Buccal mucosa was dissected from the muscle and all the layers were connected separately paying close attention to reconstruction of the orbicularis oris. The muscle was carefully dissected and sutured end-to-end, positioning the connectives lateral to the neocommissural angle. The vermilion flap was constructed on the upper lip and rotated inferiorly to join with the lower lip forming the commissure. Finally, the skin was sutured with non-absorbable single stiches (Fig. 2). Six month after the procedure the appearance was satisfactory (Fig. 3).

Conclusion

With this case report of a child with an isolated bilateral transverse cleft lip, we wanted to show how to deal with rare malformations that a surgeon will encounter only a few times in his/her career. We emphasise the importance of reviewing the literature, and when there is no gold standard for treatment, it is necessary to choose a surgical technique with which the surgeon is well acquainted. Since this is a rare malformation, there is no consensus on the general optimal surgical treatment, yet we believe that square vermilion flap combined with the end-to-end muscle repair and straight-line skin closure will give optimal results. The aetiology remains unclear but there are indications that the number of pregnancies can increase the risk of bilateral asyndromic macrostomia. Timely recognition and early surgical treatment of macrostomia have a great impact on the physical and psychological rehabilitation.

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Ethical approval

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Patient consent

Written patient consent was obtained from the parent/legal guardian.

Authors' contribution

M. Tarle and A. Tarle equally contributed to the manuscript.

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