Original Article

Tessier Number 30 Facial Cleft: A Rare Maxillofacial Anomaly

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Abstract

Introduction: In number 30 facial cleft patients, the deformities vary in their presentation, ranging from minor clefting of the lower lip (2) to complete clefts of the mandible with absence of the hyoid bone, thyroid cartilage, and manibrium. **Materials and Methods:** A case of Tessier number 30 facial cleft with only lower lip deformity is reported in this paper. **Results:** The pathognomonic features of the facial clefts were reviewed in terms of the related literature. **Conclusion:** We aimed to remind the pathology and review the literature in this report.

Keywords: Cleft lower lip, midline cleft, Tessier number 30 cleft

INTRODUCTION

Number 30 facial cleft was first reported as a caudal extension of number 14 and number 0 facial clefts by Tessier in 1976. The deformities vary in their presentation, ranging from minor clefting of the lower lip^[2] to complete clefts of the mandible with the absence of the hyoid bone, thyroid cartilage, and manibrium. In this article, we present a case of midline lower lip cleft that was repaired in late adolescent period and a review of the literature.

A 20-year-old male patient was referred to our clinic with a midline cleft and ulceration of the lower lip without any other involvement [Figure 1]. Several treatment methods such as antibiotic ointments and antihistaminic drugs had been tried for the ulcer by dermatologists; however, the pathology was not resolved.

In physical examination, the upper lip, tongue, nose, and frontal region were found normal. The mandible and the soft tissue of the neck were not involved. Radiographic confirmation was done. Written consent was obtained from the patient, and the surgical approach was planned.

Under local anesthesia, an elliptic incision was performed, ulcer was debrided, and the orbicularis oris muscle was identified. Free edges of the muscle were dissected and sutured together in the midline. Mucosa and the vermillion line were connected meticulously. No complications were observed in the early postoperative period. At 10-month follow-up, the patient was satisfied with the result [Figure 2].

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DISCUSSION

The incidence of craniofacial clefts is estimated between 1.4 and 4.9/100,000 live births.^[4] Although two genders are being affected by craniofacial clefts, girls are more commonly affected. The pathology can develop due to large doses of radiation, infection, vitamin deficiencies, metabolic syndromes, and Vitamin A toxicity.^[5-7]

In Tessier Classification, clefts 0–14 radiate around the orbital bone rims [Figure 3 and Table 1]. Tessier number 0 and number 3 are seen to be the most commonly reported cleft types in both genders in the literature. [8,9] Tessier number 30 cleft of the face is a rare congenital anomaly. The deformity occurs as insufficient fusion of first branchial arc or the failure of mesodermal penetration in midline.[10] Anomaly can range from a lower vermillion notching to the involvement of the teeth, tongue, mandible, and neck.[11-14] Tafreshi et al. reported a Tessier number 30 cleft case with congenital heart defects.^[13] One patient was reported by Adetayo and Martin^[15] with a Tessier number 30 cleft concomitant with levocardia, ventricular septal defect, a patent foramen ovale, double outlet right ventricle, intestinal malrotation, and bilateral undescended testicles. Vendramini-Pittoli et al. published two Tessier number 30 cleft cases. One of them was accompanied

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Figure 1: A 20-year-old male patient with notching and ulceration on the lower lip



Figure 2: View of the patient in the postoperative 10-month follow-up period

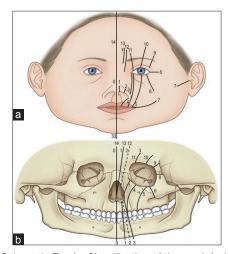


Figure 3: Schematic Tessier Classification of the craniofacial clefts (a) Bony clefts, (b) Soft tissue clefts

by a Tessier number 7 cleft, and the other one had developed together with Tessier number 4, 7, and 9 clefts.^[16] Moreover, nasal anomalies and cleft lip and palate can also be seen with Tessier number 30 facial clefts.^[17-19]

Midline cleft of the lower lip and mandible can also be a sign of a syndrome, such as the Robinow syndrome which presents itself with extremity, genitalia, costa and vertebra anomalies, [20] or the Richieri-Costa syndrome which involves limb defects. [21]

The prenatal diagnosis of the Tessier number 30 cleft can be made through ultrasonography at 15–18 weeks of the pregnancy.^[22]

Reports about this pathology are rare in the literature. [10] Less than 80 cases have been reported since the first article by Couronné. [10,23] There are a few cases of craniofacial clefts reported from Turkey; [24-27] moreover, only two papers reporting cases from Turkey that are accompanied by a simple lower lip defect. [24,28] Due to this rarity, it can be said that there is no consensus on the management of this deformity. Therefore, we decided to prepare an algorithm for the treatment of the pathology.

Simple notching of the lower lip can be treated with wedge excision or Z-plasty. [29] Lip deformity and bifid tongue should be corrected as early as possible due to functional deficits. Duplication of the tongue can be corrected with wedge excision or Z-plasty. In addition, the tongue should be released from the floor of the mouth. [30]

Clefts of the mandible can be treated with one- or two-stage procedure depending on the range of the defect. [31] Most surgeons begin the procedure when the child is about 10-year-old to avoid tooth bud damage and use interosseous wires or plates. [32] On the other hand, Oostrom *et al.* report that a carefully planned osteosynthesis of the base of the mandible does not harm the buds and may provide better occlusion. [33] If the mandibular cleft is too wide, the bones should be stabilized at an early age to avoid obstructing eating and breathing. [34] A bone graft harvested from the costa, calvarium, or iliac crest can be inserted around the age of 10 in the second stage. [28,35,36]

One of the largest case series was reported by Freitas *et al.* with 17 patients aged from 1 month to 30 years. [36] Early surgery was performed in two patients with sleep apnea and impaired nutrition using distraction osteogenesis to achieve better tongue movement. They used Z-plasty for mucosal bands, muscle repair for the contraction, and straight closure of the skin in a few patients. Early bone grafting was recommended in this report to establish mandibular continuity.

The strap muscles of the neck are often replaced with dense scar tissue and cause flexion contracture bands and can be repaired with multiple Z-plasties. [11] Facial anomalies can extend to the widened interclavicular space, bifid or absent manubrium, presternal skin tags, cleft lip and palate, hemifacial microsomia, dermoid cyst of the face, extremity anomalies, and cardiac deformities. [10] Treatment choices for all these anomalies should be determined according to the type of the deformity.

CONCLUSION

The lower lip notching was corrected with wedge resection and three-layer closure in our patient. No complications occurred. Functional and esthetic results were good in the postoperative

Table 1: Clinical features of the Tessier craniofacial clefts

Tessier classification					
	Synonyms	Major clinical features of the pathology			
Tessier number 0	Median craniofacial	Deficiency of midline structures			
	dysraphia Centrofacial microsomia Frontonasal dysplasia Median cleft face	Developmental arrest range from hypoplasia of the nasomaxillary region and hypotelorism to a severe cyclopia, ethmocephaly, or cebocephaly False median cleft lip and absence of philtral columns Narrowed columella or totally absence of columella			
	syndrome	Lack of septal support			
	Holoprosencephaly	Skeletal deficiencies range from separation between the upper central canines to absence of the premaxilla and cleft of the secondary palate Partial or total absence of nasal bones and septal cartilages			
		Hypotelorism or cyclopia			
		Encephalocele			
		Excess of midline tissue			
		True median cleft lip with broad philtral columns Duplication of the labial frenulum Bifid nose with a broad columella			
		Laterally displaced alae and upper lateral cartilages			
		Skeletal excess can be seen as a diastema between the upper central incisors			
		Duplicate nasal spine			
		Keel-shaped maxillary alveolus			
		Anterior open bite			
		Shortened central midface height			
		Cartilaginous and bony nasal septum is thickened or duplicated			
		Nasal bones and nasal process of the maxilla are broad, flattened, and displaced laterally			
		Ethmoid and sphenoid sinuses may be enlarged, contributing to symmetric widening of the anterior cranial fossa and hypertelorism Displacement of the pterygoid plates away from the midline			
Tessier number 1	Type 3 nasoschisis nasal	Soft tissue involvement			
ressier number i	dysplasia	It is similar to the common cleft lip			
	7 1	Notching on soft triangle of the nose			
		Short and broad columella			
		The nasal tip and nasal septum deviate away from the cleft			
		Malpositioned medial canthus, and telecanthus			
		Skeletal involvement			
		A keel-shaped maxilla and anterior open bite			
		An alveolar cleft that passes between the central and lateral incisors The cleft may extend posteriorly as a complete cleft of the hard and soft palate			
		The nasal bones are displaced and flattened			
Tessier number 2		Hypertelorism Soft tissue involvement			
ressiei iluliloei 2	-	Ala nasi is hypoplastic, lateral aspect of nose is flattened and dorsum is broad			
		Cleft passes medially to the palpebral fissure and eyelid is not involved Lacrimal duct is usually intact			
		Skeletal involvement			
		Cleft begins between lateral incisor and canine, extends into apertura piriformis Hard or soft palate cleft can be accompanied with the pathology			
		Cleft passes between nasal bone and frontal process of maxilla			
		Orbital hypertelorism can be occurred			
		Asymmetry of the sphenoid wings and anterior cranial base			
Tessier number 3	Oro-naso-ocular cleft	Soft tissue involvement			
		Begins like number 1 and number 2 clefts			
		Nose is shortened at the effected side			
		Cleft extends between medial canthus and lacrimal punctum, lacrimal system is affected			
		Recurrent ductal infection is common Medial canthus is displaced inferiorly, colobomas of lower eyelid are medial to inferior punctum Microphthalmia may occur, eye is malpositioned inferiorly and laterally			
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190191 Cleft Cleft Claft begins from arel commissions and extends to prequirioular hairling	Tessier number /	facial cleft					
facial cleft Cleft begins from oral commissure and extends to preauricular hairline Craniofacial microsomia Pathology can range from a mild broadening of oral commissure with a preauricular skin tag to a							
Hemifacial microsomia complete fissure extending toward the microtic ear							
Otomandibular Cleft does not extend beyond anterior border of masseter muscle							
dysostosis Ipsilateral tongue, soft palate and muscles and mastication can be affected		dysostosis					

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		Tessier classification
	Synonyms	Major clinical features of the pathology
	First and second branchial arch syndrome Auriculo- branchiogenic dysplasia Hemignathia and microtia syndrome Oro-aural cleft Group B1 lateral otocephalic branchiogenic deformity Zygotemporal dysplasia	Parotid gland and duct can be absent Facial nerve weakness may occur External ear absence or microtia or only preauricular skin tag can be seen depending on the level of pathology Middle ear can also be affected Preauricular hair is usually absent in craniofacial microsomia patients Hairy skin between oral commissure and temporal region can be seen in Teacher Collins Syndrome The ipsilateral soft palate and tongue are often hypoplastic Skeletal involvement Skeletal cleft passes through pterygomaxillary junction Cleft is centered in the region of temporozygomatic suture according to Tessier Posterior maxilla and mandibular ramus are vertically hypoplastic and cause abnormal occlusal plane Coronoid process and condyle are often hypoplastic, contributing to a posterior open bite Zygomatic body is hypoplastic and displaced In severe pathology, zygomatic arch represents as a small stump In severe form, true orbital dystopia can occur Zygomatic process of temporal bone is normal Cranial base is asymmetric and glenoid fossa is malpositioned There can be a rudimentary medial and a lateral pterygoid plate Sphenoid bone is usually abnormal
Tessier number 8	Frontozygomatic cleft The equator of the Tessier craniofacial time zones	Soft tissue involvement Cleft extends between lateral canthus and temporal region Hair markers can be seen along a line between temporal area and lateral canthus True lateral commissure coloboma with absence of lateral canthus is seen Epibulbar dermoids are also presented Skeletal involvement Cleft is located at frontozygomatic suture The only support of lateral palpebral fissure is the greater wing of the sphenoid bone
Tessier number 9	Frontosphenoid dysplasia	There is soft-tissue continuity of the orbit and temporal fossa Soft-tissue involvement Lateral third of the upper eyelid and eyebrow are abnormal Lateral canthus is distorted In severe pathology, microphthalmia is presented Superolateral bone defect causes lateral displacement of globe Cleft extends to temporoparietal hair-bearing scalp Temporal hairline is anteriorly displaced, and a temporal hair projection is often seen Nervus facialis palsy in forehead and upper eyelid is common Skeletal involvement Bone defect extends through superolateral aspect of the orbit, involving supraorbital rim and roof Upper part of greater wing of sphenoid, squamous portion of temporal bone, and surrounding parietal bones are distorted Posterolateral rotation of lateral orbital wall can be seen Pterygoid plates are hypoplastic There may be a reduction in anteroposterior dimension of anterior cranial fossa
Tessier number 10	-	Soft tissue involvement Begins at the middle third of upper eyelid and eyebrow Lateral eyebrow may angulate temporally Palpebral fissure may be elongated with an amblyopic eye displaced inferolaterally In severe pathology, entire upper eyelid can be absent Colobomas can be seen Frontal hair projection may connect temporoparietal region to lateral brow Skeletal involvement Begins in the middle of upper orbital rim just lateral to supraorbital foramen A prominent mass can be seen in forehead due to encephalocele Orbit rotated lateroinferiorly In severe disease, hypertelorism can be seen Anterior cranial base is affected

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Tessier classification						
	Synonyms	Major clinical features of the pathology				
Tessier number	-	Soft tissue involvement				
11		Medial third of upper eyelid is affected and upper eyebrow is disrupted				
		Pathology can involve a tongue-like projection of the frontal hairline				
		Skeletal involvement				
		A notch can be seen in the medial third of the supraorbital rim				
		If cleft passes through ethmoid air cells, orbital hypertelorism can be seen Cranial base and sphenoid anatomy are normal				
Tessier number	-	Soft tissue involvement				
12		Cleft lies medial to medial canthus				
		There is a lateral displacement of the canthus that causes telecanthus				
		There is no eyelid clefting				
		Aplasia of medial eyebrow can occur				
		Downward projection of paramedian frontal hairline possible, but forehead skin is normal				
		Skeletal involvement				
		Passes through frontal process of maxilla				
		Transverse dimension of ethmoid cells are increased and orbital hypertelorism occurs				
		Frontal and sphenoid sinuses are usually enlarged Cleft is lateral to olfactory groove, so cribriform plate is normal				
		Encephaloceles have not been observed in this cleft				
		Anterior and middle cranial fossae are widened				
Tessier number	_	Soft tissue involvement				
13		There is typically a paramedian encephalocele located between nasal bone and frontal process of maxilla				
		Upper eyelid and eyebrow are intact				
		Medial end of eyebrow can be displaced inferiorly				
		A V-shaped frontal hair line can be seen				
		Skeletal involvement				
		Anomaly in cribriform plate is characteristic in this cleft				
		There is widening in olfactory groove				
		Cribriform plate and ethmoid sinus are enlarged, thus hypertelorism can be seen				
		A paramedian encephalocele can relocate the cribriform plate inferiorly leading to orbital dystopia				
Tessier number	Median craniofacial	Soft tissue and skeletal tissue deficiency				
14	dysraphia	Hypotelorism can be seen				
		A spectrum of holoprosencephalic disorders such as cyclopia, ethmocephaly and cebocephaly may also be seen				
		Cranium is typically microcephalic				
		Cranial base components are absent which causes orbital fusion				
		Soft tissue excess				
		Hypertelorism can also be seen with number 14 cleft				
		Frontonasal encephalocele or a midline frontal encephalocele can cause lateral displacement of the				
		orbits				
		Forebrain remains in a low position during embryologic development				
		Glabella is flattened				
		Periorbita, eyelids and eyebrow are normal Long midline projection of frontal hairline can be seen				
		Skeletal tissue excess				
		Caudal frontal bone is flattened				
		While frontal sinus is nonpneumatized, sphenoid sinus is over-pneumatized				
		Bifid crista galli and perpendicular plate can be seen				
		Crista galli and ethmoid bone are widened and caudally displaced				
		Cribriform plate is displaced inferiorly				
		Relative shortening of middle cranial fossa is caused by rotation of sphenoid wings				
		Harlequin eye deformity can be seen on X-ray				

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Tessier classification Synonyms Major clinical features of the pathology Tessier number Mandibular process cleft Soft tissue involvement Midline branchiogenic Pathology can range from a notch in the lower lip to an entire lower lip and chin cleft syndrome Anterior tongue can be bifid and attached to split mandible Intermandibular Ankyloglossia and total absence of tongue have also been reported dysplasia Skeletal involvement Cleft is located between central incisors and extends to mandibular symphysis Anomaly is thought to be caused by failure of fusion of first branchial arch If lower branchial arches are affected, neck anomalies can be included in the pathology Hyoid bone is absent in most cases and thyroid cartilages are underformed Flexion contracture of neck due to atrophia of neck strap muscles

10-month follow-up period. Tessier number 30 cleft should be kept in mind in lower lip anomalies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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