



Surgical Management of a Rare Bilateral Tessier Number 4 and 7 Facial Cleft: A Case Report

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Abstract

Background: craniofacial clefts are rare congenital anomalies of unknown etiology of which only few case reports are available in the literature.

Case report: this is the first case report of a two-week-old child presented with bilateral facial cleft; Tessier No.4 on the right and Tessier No.7 on the left side of the face treated surgically in Kurdistan region of Iraq.

Conclusion: management of these children is complex and highly individualized, sharing more of such reports will help in developing better strategies for their treatment.

Keywords: Congenital malformation, Facial cleft, Macrostomia

Background

Facial clefts are rare disfiguring congenital malformations that affect 1.43-4.85 in 100000 births worldwide the etiology of which is unknown.¹ Craniofacial clefts were best described anatomically by Dr. Paul Tessier back in 1976 through a numerical system. Dividing them into 14 different types in relation to facial and orbital sagittal midline. Number 0-7 are counted as facial clefts and 8-14 are cranial clefts. Tessier number 4 begins between philtrum and oral commissure in the upper lip ascending

medial to the infraorbital foramen till the orbit. Abnormalities that could be observed in No.4 facial cleft are inferior displacement of medial canthus, decreased oro-ocular and oculo-alar distances, ocular dystopia, cleft lip and palate.² On the other hand Tessier facial cleft No.7 is transverse facial cleft or macrostomia that affects all the layer of the cheek mostly reaching the anterior border of the masseter muscle, Figure (1). Many surgical modalities have been recommended for correction of different facial cleft types, but none is considered superior.

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The principle of the surgical intervention is to urgently cover the exposed eye, improve feeding, speech ability and the child's appearance.³ This is the first case report of a combined Tessier No.4. & No.7 facial clefts from this region in the English literature to our knowledge and the aim is to share our experience with a literature review about its surgical management.

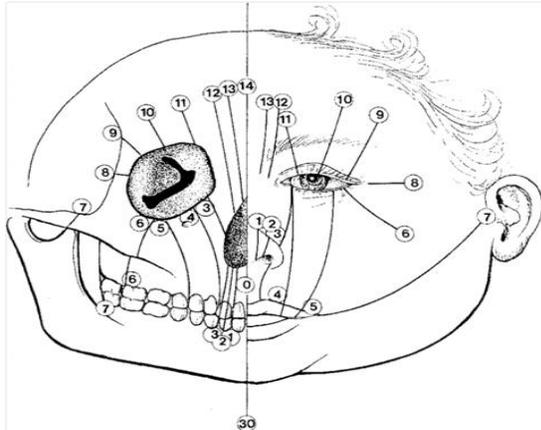


Figure (1): Tessier classification of craniofacial clefts.²

Case report

A one-week-old male child was presented to Sulaymaniyah Burn, Reconstructive and Plastic Surgery Hospital in Sulaymaniyah city Kurdistan Region-Iraq in April 2024 referred by pediatrician and ophthalmologist. The child was born to a healthy young couple who had 3 other healthy children and gave no familial history of any congenital abnormality so whatever more a significant exposure or event during the pregnancy. They were told about the facial cleft in the 22nd week of gestation and they were willing to terminate the pregnancy if it wasn't for the legislation. He had facial cleft Tessier No.4 on the right side of his face affecting all the anatomical layers of the cheek displacing the right eye and mouth with an open communication between them, the Rt. globe was exposed due to almost total absence of lower eyelid, inferior orbital floor, and maxilla. On the left side he had Tessier No.7

that reached the anterior border of masseter muscle. Without any other apparent anomaly. He had no other medical complaints besides Rt. eye exposure and feeding difficulty. Preoperative work up done to prepare him for closure of the defect and coverage of the Rt. eye. A week later under general anesthesia the Rt. facial defect was closed by a transpositional flap harvested from the medial edge of the cleft, without violating the nose, to reconstruct the anterior lamella of the lower eyelid sutured to the reflected mucosal layer that was present inferior to the globe to replace the palpebral conjunctiva, Figure (2).



Figure (2): On table photo of the child A. preoperative marking. B. postoperatively after reconstruction of the Rt. lower eyelid, Rt. Cleft lip and Lt. oral commissure





Then all the cheek in the lateral side of the cleft was undermined through supra periosteal plane and advanced superomedially to be sutured in layers to the reconstructed lower eyelid, nasal ala and lip trying to preserve as much skin as possible while tailor tucking the flaps. The cleft lip was repaired in 3 layers, mucosa, muscle and skin. Lt. Tessier No.7 reconstructed by incising both edges of the cleft then separating the anatomical layers and suturing each of them to its corresponding layer. On the next day all flaps looked well vascularized and the child was generally stable so he was sent home on oral antibacterial and pain killer medication to be seen in 1 week. Fortunately, no wound healing problems faced apart from dehiscence of a medial tarsorrhaphy stitch which was replaced under local anesthesia the same day, Figure (3).



Figure (3): one week postoperatively

A month later the child's wounds were healed well with improved feeding and the parents felt much better about his appearance, but the eye was still exposed that's why another operation was performed under general anesthesia performing lateral permanent tarsorrhaphy with medial canthopexy, Figure (4). The plan is to keep him under observation in coordination with ophthalmology and pediatrics team until the next stage of reconstruction during his preschool age.



Figure (4): one month postoperative.

Discussion

Pathophysiology of facial clefts best explained by developmental field reassignment theory stating that loss of some critical elements in the craniofacial bones formation prevents fusion and produces the defect.⁴ Some literature suggests that direct tethering of tissue migration due to amniotic band may be the cause for tissue necrosis during early gestational life resulting in these devastating craniofacial malformations.⁵ They occur in the face in different severity of involved tissues ranging from skin only to displaced or even absent eye, mouth and nose.² They may occur as isolated anomalies





or in combination with other congenital malformations and syndromes.⁶ Despite of the huge psychological impact of this disfiguring condition on the child and the parents, those children's main problem is that their eyes are usually exposed due to defective eyelids which makes them prone to vision loss and their palatal cleft cause feeding difficulty. For that the aim of the primary surgical intervention is to cover the cornea and improve feeding alongside improving appearance. There is no need to wait for skeletal maturation worrying about the chance of growth arrest as a complication because those children usually have anomalous tissue growth in the first place.⁷ The difference in severity and combination of facial clefts presentation make their management plan highly individualized and producing a reliable algorithm for their rehabilitation is not possible now because of the small number of reported cases worldwide.¹ We haven't come across any report of a similar presentation to this case having Tessier No.4 on Rt. side of the face involving soft and bony tissue with Tessier No.7 on the Lt. reaching the anterior border of Masseter muscle. Although the severity of the exact tissue involvement was not assessed because the child was only 2-week-old and no radiological study was done, Tessier No.4 can involve the maxillary bone resulting in a defect or even absence of the whole bone while sometimes only the soft tissue is affected and Tessier No.7 presentation can be highly variable affecting tongue, zygoma, maxilla and mandibular bones with or without parotid gland hypoplasia and defective cranial nerves 5 and 7 innervation.^{2,8} Interestingly we found several case reports about children with different types of facial clefts born in Iraq but were managed and reported in other countries like Turkey, India and Iran recommending studying the genetic of the population for possible mutations but this will be the first case report about a facial

cleft of this kind that was managed inside the country being bilaterally involved with two different types of Tessier clefts.⁸⁻¹¹ There are varying types of flaps used by different authors to close these facial defects depending on the availability of local tissues to transpose into the cleft without compromising the facial units ranging from simple z-plasty to expanding the forehead with tissue expander for providing sufficient skin along with bone graft to reconstruct the face.¹¹ Our surgical plan was according to the recommendations in the literature depending on autologous local soft tissue, using advancement and transpositional flaps, preserving as much skin as possible and closing the wound in layers to relieve the tension on the closure but the healing and growth of the anomalous tissues of these children are unpredictable on the long run for that he will be followed up by our multidisciplinary cleft team to address any future complaint as needed and for the appropriate timing of this palatal repair which is usually performed at 6-9months of age.

Conclusion

This report shares the surgical plan for managing a complex rare case with two different types of Tessier facial cleft treated in Kurdistan region of Iraq. We believe more of such reports would help in formulating a standard strategy for the surgical treatment and rehabilitation programs for these rare cases.

Disclosure

The authors assert that they have no conflicts of interest.

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