Original Article

# Tessier cranio-facial clefts presenting to a tertiary eye care center in Northern India: Ophthalmic features and a review of management

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**Purpose:** Tessier classification is used to classify congenital facial cleft disorders utilizing the anatomical location of the cleft and its extension. The orbital and ocular morbidities associated with the birth disorder are numerous. The authors decided to perform a retrospective analysis of the clinical features of the patients who presented to a tertiary care hospital with orbito-cranial clefts. **Methods:** The authors retrospectively evaluated the records of patients with craniofacial clefts who had presented to a tertiary eye care hospital in northern India in the last 2 years (January 2019–December 2020). The clinical features were studied, entered in MS Excel, and the data were evaluated. **Results:** The data of 40 patients with Tessier cleft were found. The majority of the patients were male and presented in the pediatric age group. Unilateral involvement was more common, with maxillary hypoplasia being the most common facial anomaly associated. Eyelid coloboma and euryblepharon was the most common periocular finding; lateral epibulbar dermoid and corneal opacity were the most common ocular surface anomaly. The majority of patients had presented for cosmetic correction. The syndromic association was with Goldenhar syndrome ($n$ = 13), Fraser ($n$ = 2), and one each of Treacher Collins, blepharocheilodontic, organoid nevus, and oculo-dento-digital syndrome. Combined clefts were also seen. **Conclusion:** Tessier cleft classification is a useful tool to classify cranio-facial left anomalies. Multitudes of ocular and orbital anomalies can be associated with their different forms. Better knowledge and understanding of the classification will aid immensely in predicting the ocular defects and planning their management.

**Key words:** Abortive cryptophthalmos, complete cryptophthalmos, eyelid developmental disorder, Fraser syndrome, syndactyly

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Cleft disorders of the head and face may be found isolated or in combination with skeletal and soft tissue disorders.[1] The condition has been studied in the past and many attempts have been made to classify them.[2,3] Tessier described the classification of orbito-cranial clefts based on their anatomic location and extension. The nomenclature included both soft tissue and bony anomalies. He used orbit as the primary reference point, described 15 different possible lines of cleft formation, and allotted a specific number to each [Fig. 1]. Numbers 0 and 14 were the median clefts, 1, 2, 12, and 13 were the paramedian clefts, 3 and 4 were the oculo-nasal cleft, 5 was the oculo-facial cleft, 6, 7, 8 were the lateral clefts, 9 was the upper lateral, 10 was the upper central, and 11 was the upper medial cleft. Although multiple case reports of individual Tessier cleft numbers exist in the literature, there are obvious large-scale studies on ophthalmic features of patients with craniofacial cleft anomalies. The author planned to perform a retrospective review of these patients who had presented to a tertiary eye care center in northern India to classify them according to the Tessier classification and study the surgical planning performed.

## Methods

This retrospective study was conducted in a tertiary eye care hospital in northern India. Medical records of all patients who presented to the oculoplasty clinic with facial cleft disorders between January 2019 and January 2020 were considered for the study. All information regarding the history, clinical features, and management was collected and entered in MS Excel and analyzed. Informed consent was obtained from all patients or their guardians.

## Results

Forty patients were identified on retrospective evaluation of institution records [Figs. 2 and 3]. The mean age of presentation was 9.73 years (range: 1 month–24 years). The ratio of males to females was 26:14, showing a higher male preponderance. Fourteen patients had bilateral and 26 patients (right: left–15:11) had unilateral Tessier cleft. Facial clefts were bilateral in 14 patients.

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Single clefts: Type 8 cleft was bilateral in three cases and unilateral in five cases. Type 10 cleft unilateral in four cases and one bilateral case had only on the left side. Type 4 cleft was noted in only five unilateral clefts. Type 3 cleft was bilateral and unilateral in two cases each, and one bilateral case had only on the left side. Only one case had unilateral type 11 cleft. One case had type 8 cleft on the right side and type 11 on the left side.

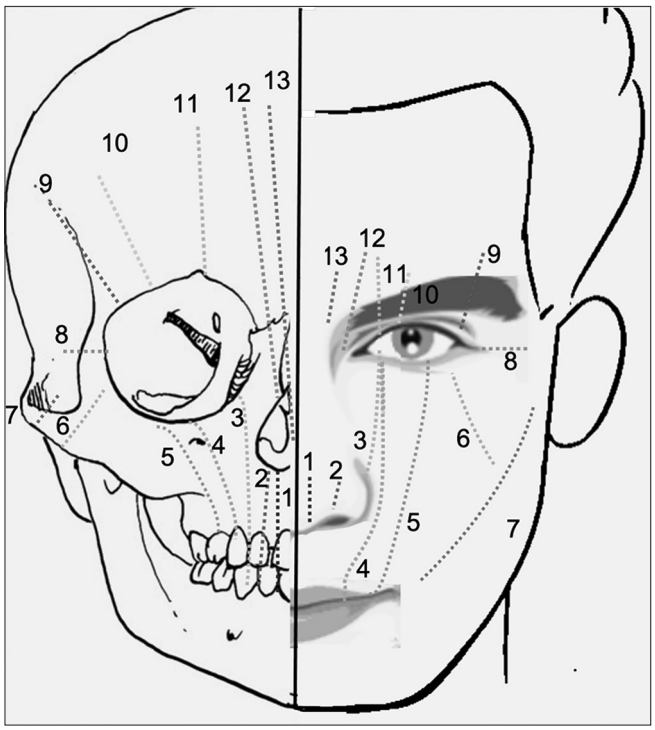


**Figure 1:** Schematic diagram showing bony (left half) and soft tissue (right half) craniofacial clefts

Combined clefts: Type 3 and 8 clefts were commonly seen (12 and 11 sides, respectively) associated with other types of clefts. The rest of the details of combined clefts are given in [Table 1].

Syndromic association was seen that included Goldenhar syndrome ($n$ = 13), Fraser syndrome ($n$ = 2), Treacher Collins syndrome ($n$ = 2), blepharocheilodontic syndrome ($n$ = 1), organoid nevus syndrome ($n$ = 1), and oculo-dento-digital dysplasia ($n$ = 1). Other associated features included hemangioma over the cheek, syndactyly, camptodactyly, oxycephaly, and arachnoid cyst.

Facial anomalies included maxillary hypoplasia ($n$ = 4), retrognathia ($n$ = 4), hemi-nasal aplasia, and hypoplasia ($n$ = 1 each), asymmetric nares ($n$ = 3), depressed nasal bridge ($n$ = 2), broad nasal bridge ($n$ = 2), microtia ($n$ = 3), and accessory auricles ($n$ = 5). Five patients had operated cleft lips during presentation [Table 2].

### Periocular anomalies

Periocular anomalies included upper eyelid coloboma ($n$ = 17), lower eyelid coloboma ($n$ = 12), euryblepharon ($n$ = 12), eyebrow madarosis ($n$ = 9), ptosis ($n$ = 1), lateral canthal dystopia ($n$ = 1), medial canthal dystopia ($n$ = 2), ectropion ($n$ = 1), and synorphys ($n$ = 1). Lacrimal drainage system anomalies included nasolacrimal duct obstruction ($n$ = 7), punctal agenesis ($n$ = 10), absent lacrimal sac ($n$ = 4), and displaced punctum ($n$ = 6). Orbital rim notching was noted superiorly ($n$ = 5), superotemporally ($n$ = 2), inferonasally ($n$ = 14), and inferotemporally ($n$ = 1).

### Ocular surface anomalies

Limitation of elevation ($n$ = 1), adduction ($n$ = 1), and depression ($n$ = 1) on examination for extraocular motility. Anterior segment anomalies included lateral epibulbar

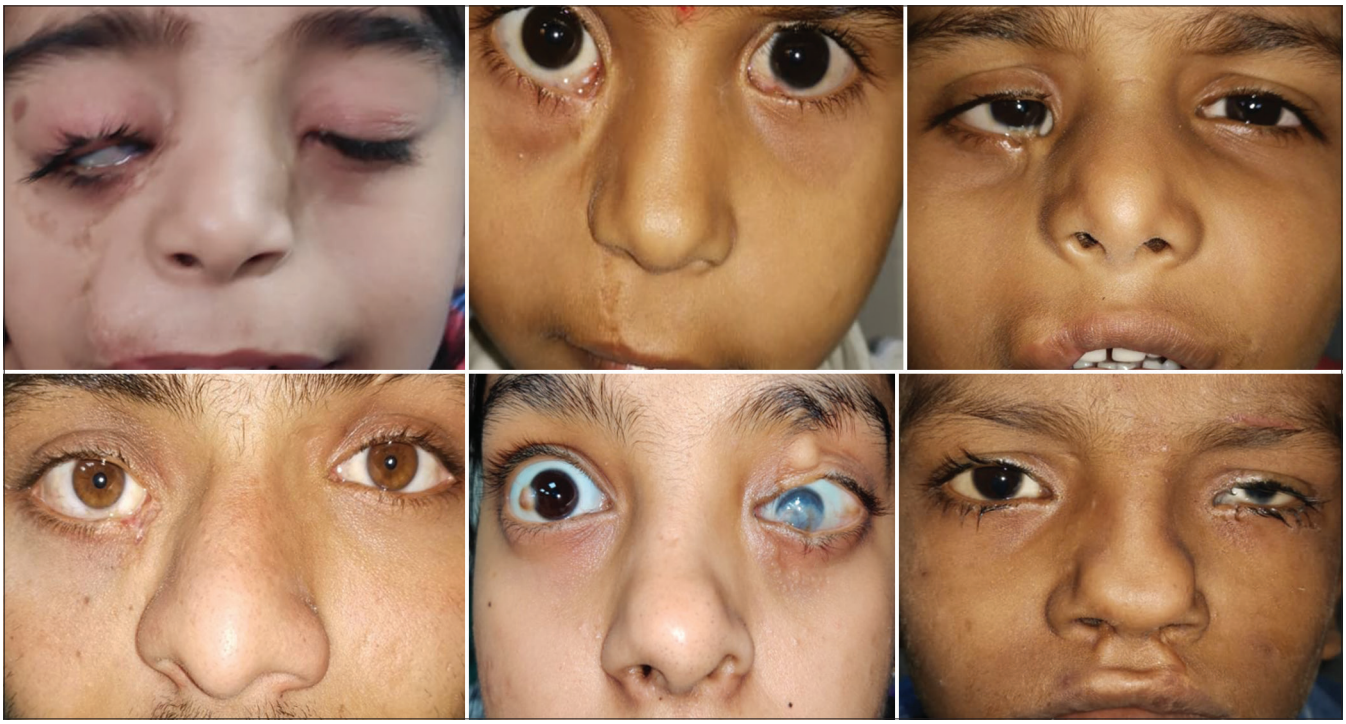

**Figure 2:** Clinical image of patients with unilateral Tessier cleft

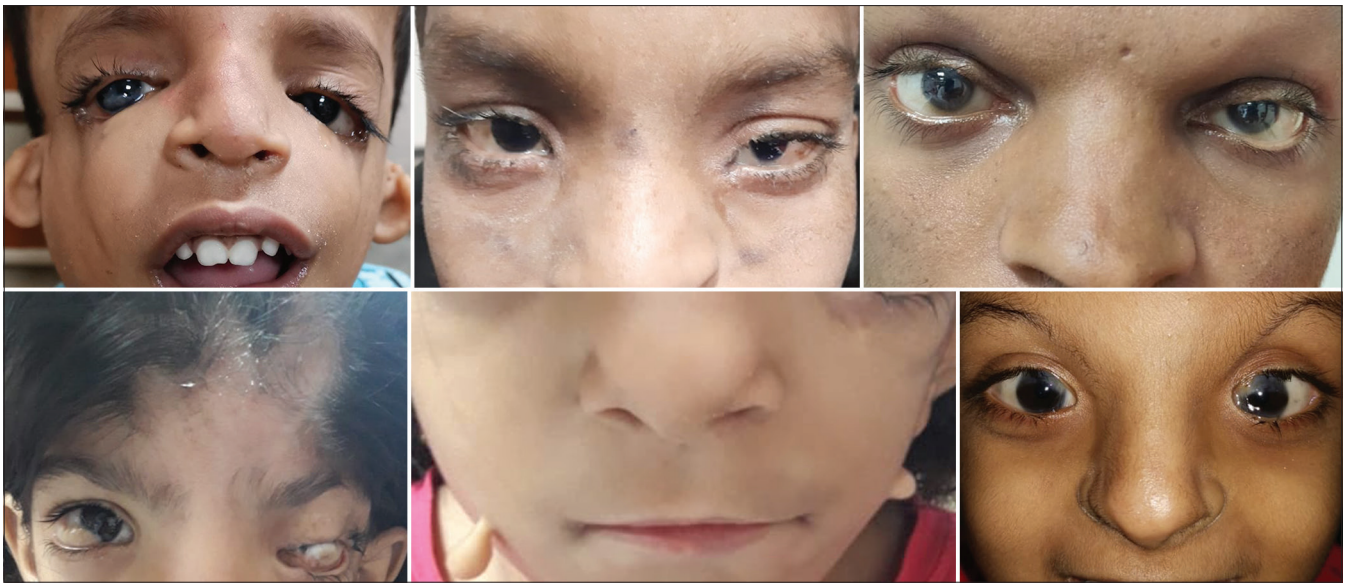

**Figure 3:** Clinical image of patients with bilateral Tessier cleft

**Table 1: Features of combined clefts and their common combinations**

| Tessier number | Ophthalmic features |
|---|---|
| 0,1,2,14 | Hypertelorism, microphthalmos |
| 3,4 | Lower eyelid coloboma<br>Lacrimal drainage anomalies<br>Symblepharon<br>Microphthalmos<br>Irido-fundal coloboma |
| 5 | Lower eyelid coloboma<br>Symblepharon<br>Lagophthalmos |
| 6,7,8 | Euryblepharon<br>Lateral canthal dystopia<br>Lipodermoid |
| 9,10,11,12 | Eyebrow madarosis<br>Eyebrow cleft<br>Eyelid coloboma<br>Superior symblepharon<br>Anterior staphyloma |

dermoid ($n$ = 8), corneal opacity ($n$ = 9), localized limbal stem cell deficiency ($n$ = 4), limbal dermoid ($n$ = 2), microcornea with iris coloboma ($n$ = 6), microphthalmos ($n$ = 3), superior symblepharon ($n$ = 3), inferior symblepharon ($n$ = 4), anterior staphyloma ($n$ = 1), and complex choristoma ($n$ = 1). Exposure keratopathy with conjunctival keratinization was present in seven patients due to lagophthalmos. On fundus examination of 30 patients, fundal coloboma was noted in six patients.

### Surgery

The majority of patients had presented for cosmetic correction due to the presence of dermoid, coloboma, canthal dystopia, euryblepharon, symblepharon, and ectropion, which were managed by performing excision ($n$ = 8), medial ($n$ = 5), or lateral ($n$ = 2) canthoplasty, direct closure of coloboma ($n$ = 4), or by skin grafting ($n$ = 2) and symblepharon release with amniotic membrane graft. Patients having lagophthalmos causing exposure keratopathy changes had undergone lateral tarsorrhaphy ($n$ = 2), skin grafting ($n$ = 1), and direct closure of coloboma ($n$ = 1). Dacryocystorhinostomy ($n$ = 5) was performed in patients complaining of epiphora.

## Discussion

The first description of cranio-facial cleft was given by Albretch in 1885, after which Morian described three types of facial coloboma in 1887.[4] Burian attempted to classify craniofacial defects in 1953; however, a proper anatomic classification was first suggested by Gorlin in 1970.[4] All confusing terminologies were eventually removed by Tessier as he had described his nomenclature for cranio-facial clefts to build a tridimensional understanding of cranio-facial malformations.

Cranio-facial, orbito-maxillary clefts, and lateral facial clefts are rare clefts compared to cleft lip disorder,[4] the exact incidence of these cranio-facial clefts is not clearly known; however, a few studies estimate it to be between 1.4 and 6 per 100,000 live births.[5,6]

In a retrospective study by Bello *et al.*,[7] the authors noted that Tessier 1 was the commonest of all clefts (24%). There were 35 (60.3%) cases of middle cleft, 14 (24.2%) cases of oblique cleft, and 9 (15.5%) cases of the lateral cleft. Typical cleft lip and palate coexisted with atypical facial cleft in two (5.6%) patients. The cleft was found to be median in 12 (33.3%) patients and right-sided in 9 (25%) patients. However, in our study, the most common cleft was type 8 and type 3.

Ophthalmic features in these clefts are multiple; however, we observed that each Tessier cleft number had a unique constellation of features. The ocular and orbital findings differ when the clefts occur isolated and when they are combined.

### Ocular findings in isolated clefts

Hypertelorism is the prevalent ophthalmic feature in cases of median and paramedian Tessier clefts (0,1,2,14,13,15).[8,9] Our retrospective study did not reveal any case fitting to median clefts. The possible reason is that these clefts are mostly associated with encephalocele and have fewer ocular abnormalities usually reported to the maxillofacial surgery department instead of ophthalmology.

**Table 2: Clinical details of patients**

| Case no. | Age/ sex | Tessier cleft | | Syndromic association | Facial features | Other features | Periocular features | Lacrimal system | Ocular surface and other findings | Presenting complaint | Management advised |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | OD | OS | | | | | | | | |
| 1 | 14/M | 8 | - | GS | Retrognathia, Broad philtrum | - | Euryblepharon, | NA | Lateral epibulbar dermoid | Cosmetic | Dermoid excision |
| 2 | 12/M | - | 3,4 | - | - | - | LL Col, IN OR notch | Displaced lower punctum | Inferior symblepharon, Localised LSCD | Epiphora, Lagophthalmos and Cosmetic | Medial canthoplasty |
| 3 | 4 m/F | 10 | - | - | Cheek hemangioma | Operated cleft lip and palate | UL Col., Madarosis, Superior OR notch | Displaced upper puncta | Superior symblepharon | Lagophthalmos | Direct closure |
| 4 | 6/F | 4,6 | 3 | GS | - | Operated cleft lip and palate | LL Col., IN OR notch | - | Microcornea with IFC | Lagophthalmos | Skin grafting |
| 5 | 1/M | - | 3,8 | - | Heminasal hypoplasia | - | UL shortening, IN OR notch | Punctal agenesis | Exposure keratopathy, conjunctival keratinization | Lagophthalmos | Lateral tarsorrhaphy |
| 6 | 11/M | - | 8 | - | - | - | Euryblepharon, Lateral canthal dystopia, UL Col. | NA | Conjunctival keratinization | Cosmetic | Lateral canthoplasty |
| 7 | 19/M | 3,4 | - | - | - | - | LL Col., IN OR notch | Lower punctal agenesis | NA | Cosmetic | Medial canthoplasty |
| 8 | 12/M | 3,8 | - | - | - | - | Euryblepharon | NA | Lateral epibulbar dermoid | Cosmetic | Dermoid excision |
| 9 | 11/F | 3,10 | - | GS | Asymmetric nares | Syndactyly | UL Col., Madarosis, Superior OR notch | Punctal agenesis | Corneal opacity with conjunctival keratinization | Cosmetic | Skin grafting |
| 10 | 11/M | 8 | 8 | FS | - | Operated cleft lip | Euryblepharon | NA | NA | Cosmetic | Lateral canthoplasty |
| 11 | 3 m/M | 8,10 | 8 | TCS | - | - | UL Col., Euryblepharon | Displaced upper puncta, NLDO | Corneal opacity, limbal dermoid, conjunctival keratinization | Lagophthalmos | Direct closure |
| 12 | 11/M | 10 | - | GS | - | - | UL Col., Madarosis, ST OR notch | Displaced upper puncta | NA | Cosmetic | Dermoid excision with coloboma repair |
| 13 | 7/M | 6,7,8 | 6,7,8 | - | Microtia | Oxycephaly | LL Col., IT OR notch | NA | Corneal opacity | Cosmetic | Lubricants |
| 14 | 8/M | 4 | - | TCS | - | - | LL Col., IN OR notch | Lower punctal agenesis | Inferior symblepharon, localized LSCD | Cosmetic | Lubricants |
| 15 | 21/M | 4 | - | - | - | - | LL Col., IN OR notch | Lower punctal agenesis | Localized LSCD | Cosmetic | Medial canthoplasty |

*Contd...*

**Table 2: Contd...**

| Case no. | Age/ sex | Tessier cleft | | Syndromic association | Facial features | Other features | Periocular features | Lacrimal system | Ocular surface and other findings | Presenting complaint | Management advised |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | OD | OS | | | | | | | | |
| 16 | 2/M | 10 | - | - | - | - | UL Col., Madarosis, Superior OR notch | Displaced upper puncta | Corneal opacity, localized LSCD | Lagophthalmos | Tarsorrhaphy |
| 17 | 2/F | 8 | 8 | GS | Accessory auricles | - | Euryblepharon | | Limbal dermoid | Cosmetic | Dermoid excision |
| 18 | 23/F | 3,8 | 3,8 | GS | Maxillary hypoplasia | - | Euryblepharon | | Limbal dermoid | Cosmetic | Dermoid excision |
| 19 | 2 m/F | 3,8,9 | 10 | GS | Accessory auricles, Microtia, Intranasal mass | Operated cleft lip | UL Col., Euryblepharon, Superior OR notch | Punctal agenesis, absent sac | Anterior staphyloma, Corneal opacity | Lagophthalmos | Dermoid excision with coloboma repair with Tarsorrhaphy |
| 20 | 23/F | 6,8 | 6,8 | GS | Accessory auricles, Retrognathia | - | Euryblepharon | NA | Lateral epibulbar dermoid | Cosmetic | Dermoid excision |
| 21 | 7/F | 8 | 8 | GS | Accessory auricles, Cheek skin tags, Depressed nasal bridge | High arched palate | Euryblepharon, UL Col., Madarosis (OS) | NA | Lateral epibulbar dermoid | Cosmetic | Dermoid excision |
| 22 | 8/M | - | 4 | BCD | Retrognathia | - | Ectropion, IN OR notch | Lower puncta not apposed to the globe | Symblepharon, Conjunctival keratinization, Corneal opacity, MED | Cosmetic | Skin grafting |
| 23 | 8/F | 8 | 11 | GS | Microtia | - | Euryblepharon, UL Col. | NA | Lateral epibulbar dermoid | Cosmetic | Direct closure |
| 24 | 2/M | - | 8 | GS | Accessory auricles | - | Euryblepharon | NA | Lateral epibulbar dermoid | Cosmetic | Dermoid excision |
| 25 | 9/M | 3 | 3 | - | Depressed nasal bridge | Operated cleft lip | NA | NLDO | Microcornea with IFC (OS) | Watering and discharge | DCR |
| 26 | 9/M | 3,11 | 3,11 | FS | - | - | UL Col. (shallow) | NLDO | Corneal opacity | Watering and discharge | Topical antibiotics |
| 27 | 15/F | - | 3,4 | - | - | - | LL Col., IN OR notch | Punctal agenesis, NLDO | Microphthalmia with IFC | Cosmetic | Medial canthoplasty |
| 28 | 24/F | - | 11 | GS | Asymmetric nares, cheek skin tag | - | UL Col., Superior OR notch | | Corneal opacity | Cosmetic | Skin graft |
| 29 | 6 m/M | - | 11,10 | ONS | - | - | UL Col., | | Choristoma | Cosmetic | Coloboma repair |
| 30 | 2/M | - | 11,8 | - | - | - | UL Col., | Displaced upper punctum | Superior symblepharon | Cosmetic | Coloboma repair with therapeutic syringing |

*Contd...*

**Table 2: Contd...**

| Case no. | Age/ sex | Tessier cleft | | Syndromic association | Facial features | Other features | Periocular features | Lacrimal system | Ocular surface and other findings | Presenting complaint | Management advised |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | OD | OS | | | | | | | | |
| 31 | 24/M | - | 4 | - | - | - | LL Col., IN OR notch | NLDO | Iris Coloboma | Cosmetic | Medial canthoplasty |
| 32 | 22/M | 3 | - | - | - | High arched palate | LL Col., IN OR notch | NA | Microphthalmia with cyst | Cosmetic | Medial canthoplasty |
| 33 | 7/M | 3 | - | - | Asymmetric nares | - | LL Col., Ptosis, IN OR notch | NLDO | NA | Watering and discharge | DCR |
| 34 | 23/M | 9,10,11 | 9,10,11 | - | Broad nasal bridge | - | UL Col., Madarosis | NA | Superior symblepharon | Cosmetic | Symblepharon release |
| 35 | 10/M | 3 | 3 | ODD | Broad nasal bridge, Asymmetric nares, Maxillary hypoplasia | Camptodactyly, High arched palate | Medial canthal dystopia, IN OR notch | NLDO, dacryocele | Microcornea with IFC | Watering and discharge | DCR |
| 36 | 6/M | 10 | - | - | Retrognathia | Arachnoid cyst | UL Col., Madarosis, ST cleft | NA | Exposure keratopathy with conjunctival keratinization | Watering and discharge | Lubricants |
| 37 | 13/M | - | 1,2,3 | - | Heminasal aplasia, Maxillary apoplasia | - | Medial canthal dystopia, Synorphys, IN OR notch | Absent sac | Microphthalmos | Watering and discharge | DCR |
| 38 | 1/F | 8 | - | GS | - | - | Euryblepharon | NA | Lateral epibulbar dermoid, Abduction limitation | Cosmetic | Dermoid excision |
| 39 | 1 m/M | 10 | - | - | - | - | UL Col., Madarosis | NA | Corneal opacity with superior symblepharon, Depression limitation | Lagophthalmos | Lubricants (planned for Cutler beard flap) |
| 40 | 4/F | 4 | - | - | Maxillary hypoplasia | Operated cleft lip and palate | LL Col., IN OR notch | Punctal agenesis | Inferior conjunctival scarring | Watering and discharge | Lubricants |

GHS- Goldenhar syndrome, FS- Fraser syndrome, TCS- Treacher Collins syndrome, BCD- Blepharocheilodontic syndrome, ONS- Organoid Nevus syndrome, ODD- Oculo-Dento-Digital syndrome, LSCD- Limbal stem cell deficiency, OR- Orbital Rim, IN- Infero nasal, IT- Infero temporal, SN- Supero nasal, UL- upper eyelid, LL- lower eyelid, Col. - Coloboma, IFC- iridofundal coloboma, MED- monoocular elevation deficit, DCR- Dacryocystorhinostomy, NA- no abnormality detected

In cases of numbers 3 and 4, the ophthalmic features include infero-medial lower eyelid coloboma, lacrimal drainage anomalies, and a symblepharon originating in the infero-medial aspect with surface keratinization, microphthalmos, anophthalmos, microcornea, lenticular coloboma, and irido-fundal coloboma.[10-12]

Tessier cleft 5 has a cleft running from the lower eyelid to the midface, the ocular features generally include microphthalmos, irido-fundal coloboma, shortened lower eyelid, or lower eyelid coloboma resulting in lagophthalmos, ocular surface keratinization, and underlying notch in the inferior orbital rim.[13]

Tessier clefts 6, 7, and 8 usually occur in combination. In our study, we found a case of isolated Tessier 6; the ophthalmological features noted in that case were lower eyelid coloboma, lagophthalmos, and exposure keratopathy. Tessier 8 may be seen isolated.[1] The ophthalmic features include euryblepharon, lateral canthal dystopia, and most of the time a choristoma is seen on the lateral bulbar conjunctiva.

Tessier 9, 10, 11, and 12 are clefts involving the superior aspect of the orbit. The ophthalmic features include eyebrow madarosis, abnormal hairline, upper eyelid coloboma, superior symblepharon, ocular surface keratinization, and anterior staphyloma.[14,15] In our study, we found that the madarosis was lateral in cleft 9 and medial in cleft 12.

### Ocular findings in combined clefts

Tessier clefts have been reported to occur in combination numerous times in the past. The clefts lying in proximity tend to occur together. The median and paramedian Tessier 0, 3, and 4 have been reported in the past; so are reports of Tessier 6, 7, and 8, and the combination of Tessier 9, 10, and 11.[16] The ocular features in such scenarios are a mixture of ophthalmic findings of both.

### Management of Tessier clefts

The management of ophthalmic features also depends on the ocular disorders. Lacrimal drainage system anomalies in Tessier 3 and 4 can be surgically corrected by performing a dacryocystorhinostomy or conjunctivo-dacryocystorhinostomy depending on the patency of the canalicular system. The infero-medial eyelid coloboma can be managed with the help of a medial canthoplasty or freshening of edges and attempting direct closure.

In Tessier 5, the surgical planning depends on the amount of anterior lamellar shortening and the severity of lagophthalmos. In mild cases, no surgical intervention is required; however, severe cases may warrant the use of full-thickness skin grafting.

The lateral canthal clefts, namely Tessier 6 and 8 can be surgically managed by performing a lateral canthoplasty or lateral tarsorrhaphy, tailoring the surgery as per the extent of euryblepharon. The often-associated lipodermoid should be carefully excised, taking care not to damage the underlying lateral rectus muscle or the palpebral lobe of the lacrimal gland. The wound can be closed in an amniotic membrane graft if required.

Surgical management of the superior group of clefts 9, 10, 11, and 12 includes full-thickness skin grafting for anterior lamellar shortening, lid sharing procedures for colobomas, and symblepharon release with amniotic membrane grafting, and fornix formation in cases of symblepharon.

Ocular surface disorders should be managed with copious lubricants. Counseling the parents regarding the disease and explaining the necessity of multiple surgeries is a must in all the above cases.

## Conclusion

Each Tessier cleft number has a characteristic group of ophthalmic disorders, identifying the number helps in predicting the exact nature and extent of the pathology. The most common Tessier cleft presenting to the ophthalmology department is Tessier numbers 3 and 8, which are different from those presenting to the maxilla-facial surgery or otorhinolaryngology department. The management of these should be planned carefully taking into account all the surrounding abnormalities.

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### Conflicts of interest

There are no conflicts of interest.

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