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Congenital Lacrimal Fistula-Dacryo-computed tomography as a diagnostic technique

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Abstract
We report the case of a 2-year-old child who presented with watering and bilateral lacrimal fistulae. Diagnosis was confirmed by Dacryo-Computed tomography. The child underwent surgical treatment for the same. This is the first reported pediatric case with bilateral congenital lacrimal fistula where Dacryo-Computed tomography was used as a diagnostic tool which aided in visualizing the lacrimal fistula and in planning for the precise surgical management. Our case report can act as a reference for clinicians in the management of bilateral congenital lacrimal fistula.

Keywords: Dacryo-Computed tomography, congenital lacrimal fistula, nasolacrimal duct

Introduction
Congenital lacrimal fistulae are rare developmental anomaly. They are accessory ducts that connect the lacrimal drainage system (lacrimal sac, the common canaliculus or nasolacrimal duct) to the skin. They are usually asymptomatic and are frequently missed due to their small size and lack of skin pigmentation making them relatively inconspicuous. Sometimes, they may be accidentally noticed on investigations like Dacryo-Computed tomography or intra-operatively [1].

Congenital lacrimal fistula represents rare developmental anomaly, the exact prevalence is unknown. Lacrimal fistulae may have either autosomal-dominant or autosomal-recessive inheritance or may be part of a syndrome. They are frequently unilateral, although familial cases tend to be bilateral [3]. The majority of these fistulae originate from the common canaliculus, but in some cases they may arise from the lacrimal sac [4].

Ever since congenital lacrimal fistulae were first described, there have been multiple reports on this condition and various methods have been used in diagnosing and treating congenital lacrimal fistula. In this case report, we will discuss the clinical presentation, investigations, and management of congenital lacrimal fistula. It is mandatory to screen patients with congenital lacrimal fistula for underlying systemic anomalies.

Case report
A 2-year-old boy was referred by the pediatrician to the ophthalmology clinic for tearing from both eyes and pits on both sides of the nose. He is the sixth child of a non-consanguineous marriage, and had a full-term normal delivery with normal milestones. The parents gave a history of discovering bilateral pits located just below the eyes on either side of the nose. There was no history of drainage or infection, or any color changes around the pits.

Ophthalmic examination revealed bilateral pigmented pits. The opening was located inferiorly and medial to the medial canthi, they were less than 1 mm in size overlying the lacrimal sac on both sides with no evidence of a complete canalization (Figure 1). Lacrimal-cutaneous fistula was found on trans illumination over the lacrimal fistula**.** There was a small amount of sebaceous crust found in the depth of the pits with fine hair at the margin of the pit.

There was no regurgitation from both puncta when pressure was applied to the lacrimal sacs. Normal tear meniscus was observed. The rest of the ophthalmic examination was unremarkable. A Dacryo-Computed tomographic imaging was obtained in order to confirm the diagnosis of bilateral congenital lacrimal fistulae as shown in figure 2&3. No other systemic, nasal, or ocular anomalies were found. No other family members were found to have lacrimal fistulae. The patient was referred to the pediatrician to rule out associated systemic abnormalities.

Fig 1: Lacrimal fistula opening inferior and medial to the medial canthi

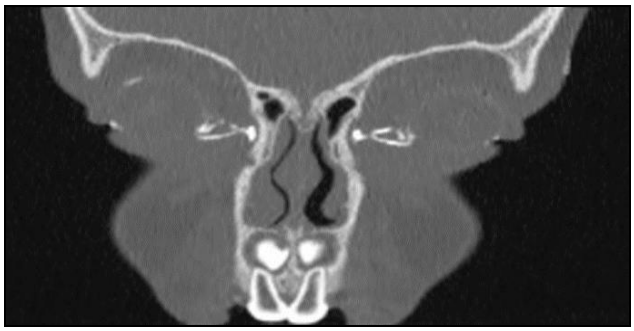

Fig 2: A Dacryo-Computed tomography of the naso-lacrimal duct coronal view showing bilateral fistula as indicated by yellow arrows

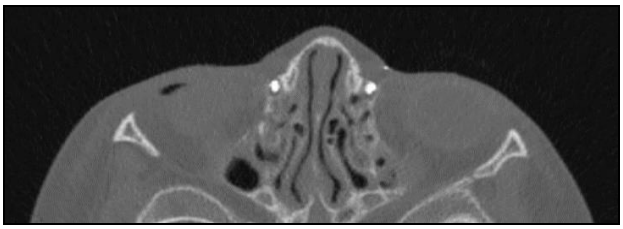

Fig 3: A Dacryo-Computed tomography of the naso-lacrimal duct axial view showing bilateral fistula.

The child underwent bilateral fistulectomy with lacrimal intubation. Intraoperatively Methylene blue was used to trace the fistula tract for complete removal of the epithelium. Patient was followed up post-operatively and silicon tube was removed after 6 weeks.

The excised tissue was sent for histopathology. The slides revealed a duct structure covered by a keratinized squamous epithelium in the superficial part and stratified squamous epithelium in the deeper part. The stratified squamous epithelium suggests canalicular origin of the fistula. A small lymphoid aggregate is seen around the ducts. No active inflammatory infiltrate seen as shown in Figure 4.

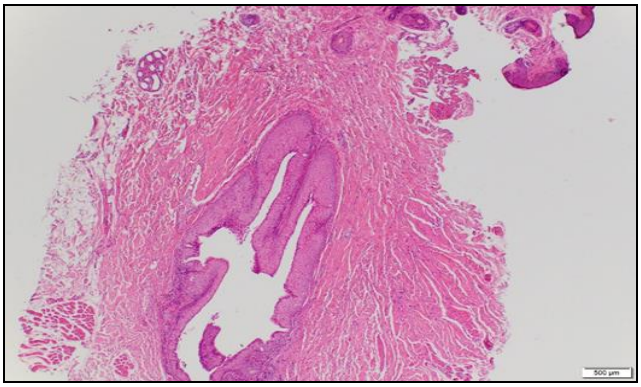

Fig 4: Histopathology specimen from lacrimal fistula showing stratified squamous epithelium suggesting canalicular origin of the fistula. The yellow arrow indicates the duct lumen.

Discussion

The lacrimal drainage system starts developing as an ectoderm thickening in the naso-optic fissure at the 7-mm stage. Eventually, this ectodermal tissue gets embedded in the mesenchyme, canalizes, and grows in the cephalic and caudal directions forming the canaliculi and nasolacrimal duct respectively. Canalization begins around the eighth week of gestation and continues until birth [1, 2].

There are different theories describing the etiopathogenesis of congenital lacrimal fistula. The well-accepted hypothesis is backed by histopathological analysis proposed by Welham *et al*. where the fistula was evident to be originating as an out-growth from the common canaliculus. However, the pathogenesis of lacrimal sac fistula can involve both the failure of fusion combined with aberrant budding from the lacrimal sac. Generally, accessory ducts form when lacrimal cells proliferate and canalize instead of involuting [1].

Congenital lacrimal fistulae are frequently asymptomatic. Symptomatic cases may present at birth with chronic epiphora or mucoid discharge from the fistula or the eye depending on the association of either transient functional or permanent anatomical nasolacrimal duct obstruction. There may be late-onset epiphora from underlying intermittent functional nasolacrimal duct obstruction. Patients may also be noticed to have clear discharge from the fistula site when coughing or blowing the nose [6]. The fistulae typically have an opening along the skin surface located inferonasal to the medial canthal angle [1].

A fullness can be felt on palpation over the region of the lacrimal sac signifying a mucocele when associated with a nasolacrimal duct obstruction [1]. Congenital lacrimal fistula may be associated with other lacrimal system anomalies like lacrimal agenesis, incomplete punctal canalization, duplicated canaliculus, nasolacrimal duct obstruction or lacrimal tract stenosis and accessory puncta. However, when associated with craniofacial clefting syndrome (e.g., Goldenhar Syndrome, charge Syndrome), intraocular congenital malformations or eyelid colobomas may also be present [6]. Multiple reports have described the co-existence of congenital lacrimal fistula with various systemic conditions like Down syndrome, naso-orbital meningocele, VACTERL associations (vertebral anomalies, anal atresia, cardiac malformations, tracheo-esophageal fistula, renal anomalies, and limb anomalies), and CHARGE syndrome (Coloboma, heart defect, atresia choanae, retarded growth and development, genital hypoplasia, ear anomalies) [8].

Standard methods of diagnostic investigations of the nasolacrimal duct anomalies include probing, irrigation and radiological methods such as a dacryocystography or nuclear scintigraphy [9, 10]. There is no gold standard for diagnosing congenital lacrimal fistula in a pediatric patient. We are highlighting the importance of Dacryo-Computed tomography in visualizing the anatomy of the nasolacrimal duct, and the detection of lacrimal fistula, in addition to the exclusion of other anomalies.

Conservative treatment is generally reserved for uncomplicated and asymptomatic congenital lacrimal fistulae especially when not associated with nasolacrimal duct obstruction.

Treatment modalities for symptomatic fistula discussed in the literature reviewed include methods such as nasolacrimal duct probing, cauterization of the external ostium, and fistulectomy with or without dacryocystorhinostomy [7]. Controversy surrounding the

efficacy of dacryocystorhinostomy in the treatment of lacrimal fistulae remains [5].

Conclusion

Our case is exclusive as the diagnosis was confirmed by Dacryo-Computed tomography. The child underwent surgical treatment for the same. This is the first reported pediatric case where Dacryo-Computed tomography was used as a diagnostic tool which aided in visualizing the lacrimal fistula and in planning for the precise surgical management. Our case report can act as a reference for clinicians in the management bilateral congenital lacrimal fistula.

Conflict of Interest

Not available

Financial Support

Not available

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