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Ectopic accessory parotid system: A rare case report

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Abstract
The ectopic accessory parotid system (EAPS) refers to a specific anomaly in the development of the parotid salivary system that is characterized as a congenital saliva-draining fistula near the oral commissure. The presence of an accessory parotid gland with an ectopic fistulous duct is a rare occurrence. We present a case of 8-year-old female child with the complaint of watery discharge from right side of the cheek. Clinical examination demonstrated a small punctum on the right side of the face, ipsilateral preauricular appendages. The diagnosis was based upon the ultrasonography, contrast digital fistulography, contrast CT sialography. Which confirmed the diagnosis of Ectopic Accessory Parotid System with cheek fistula. It was managed surgically under general anaesthesia by trans- positioning of the duct to open into the buccal mucosa near 1st molar region using Delore's method and excision of preauricular appendage on the same setting.

Keywords: ectopic accessory parotid system (EAPS), accessory parotid gland, cheek fistula, oculo-auriculovertebral spectrum (OAVS)

1. Introduction
Parotid fistulas are of acquired type and arise secondary to trauma, surgery, malignant tumours and inflammation. Rarely do they occur congenitally as malformation during foetal development. Although accessory parotid gland is a common variation, parotid fistula arising from an ectopic accessory parotid gland is extremely rare [1]. The site of opening of fistula is near the oral commissure [2]. We report a case of Ectopic Accessory Parotid System (EAPS).

2. Case report
A 08-year-old female child reported with the complaint of watery discharge from an opening in the right cheek. The flow was abundant during food intake. Clinical examination demonstrated a small punctum on the right side of the face, at a distance of about 2.4 cm from the angle of the mouth and 1cm inferior to a line joining the tragus and angle of mouth. Expression of clear serous fluid on lemon challenge test was seen. On regional examination, auricular appendage just anterior to the tragus was noted on the right side. (Figure 1)
As a part of the diagnostic work up, ultrasonography, contrast digital fistulography, contrast CT sialography was performed. These investigations revealed an isodense lobulated mass like lesion seen within the right buccal space abutting buccinator muscle anterior to right masseter within the line of right parotid duct. No communication between the accessory duct and the main duct was seen. Investigations confirmed the clinical diagnosis of ectopic accessory parotid system with cheek fistula. (Figure 2)
Intraoral trans-positioning of duct was planned using Delore's method under general anaesthesia. Ductal opening was canulated up to 1 to 1.5 cm approximately and & methylene blue dye was injected. Elliptical incision was given around the opening. Further, Duct was tunnelled, re-routed and sutured intra-orally (figure 3). Aural appendage was excised and sutured. Patient is kept under follow-up for3 months and doesn't show any signs of recurrence with good intraoral flow of the saliva.

3. Discussion
The EAPS denotes a discrete, independent salivary drainage pathway composed of a functional salivary gland outside the parotid area and its duct, which drains saliva externally through a pit near the oral commissure, but in a rare case the ductal opening was seen intraorally. Embryologically, the event of the EAPS is poorly understood. During the fourth week of embryonic development, the ectodermal lining of the stomodeum gives rise to buds or branches which form solid cords with round ends, which subsequently grows into ducts and acini. Whereas accessory salivary gland is formed from aberrant buds which lose their communication with the main salivary gland [3]. Only sixteen cases have been reported in the literature till date.
Clinically, it presents as saliva, draining through pit near the oral

commissure. Its association with preauricular appendages and occasional mandibular hypoplasia links it to craniofacial dysmorphogenesis. Accordingly, EAPS presently is taken into account within the oculo-auriculovertebral spectrum (OAVS) jointly of its milder prototypes [4].

The OAVS, which is characterized by hemifacial microsomia, first and second pharyngeal arch anomalies are characterized by multisystem involvement. Fistulae deriving from abnormal development of an adjunct salivary gland in patients with OAVS are rare. It can have a myriad of phenotypic presentation involving auricular defects (pre auricular swellings, microtia, hearing loss) 83%, facial defects (mandibular hypoplasia, hemfacia lmicrosomia, salivary fistula, cleft lip and palate) 65-75%, ocular defects (microphthalmia, epibulbardermoids etc.) 66% [5, 6]. Beleza- Meireles *et al*., have even proposed that "Isolated hemifacial microsomia with a family history of OAVS" is a stand-alone criterion for diagnosing OAVS [7]. Since, EAPS is almost universally existing with preauricular appendages and can also additionally have ipsilateral mandibular hypoplasia, their inclusion within the domain of OAVS is justified. A congenital saliva-draining cheek fistula with preauricular appendage is suggestive of EAPS.

Various methods of management of EAPS are superficial parotidectomy, intraoral transpositioning of the accessory duct, chemical cauterization. [8] and Ligation of the fistulous tract. Yet transposition of the tract is a simple procedure as described and it avoids the morbidity and radicalness of gland excision.

EAPS is a rare congenital anomaly which presents as a extraoral saliva draining fistula near oral commissure. It is invariably associated with other congenital anomaly such as preauricular appendages, mandibular hypoplasia and other craniofacial dysmorphogenesis, which is suggestive of developmental aberration of the first and second pharyngeal arch, which can be considered as a part of OAVS, though genetic basis of this syndromic disorders is yet to be established. Though various treatment options have been tried, most successful treatment has been intraoral surgical transpositioning of the duct as reported in the literature.

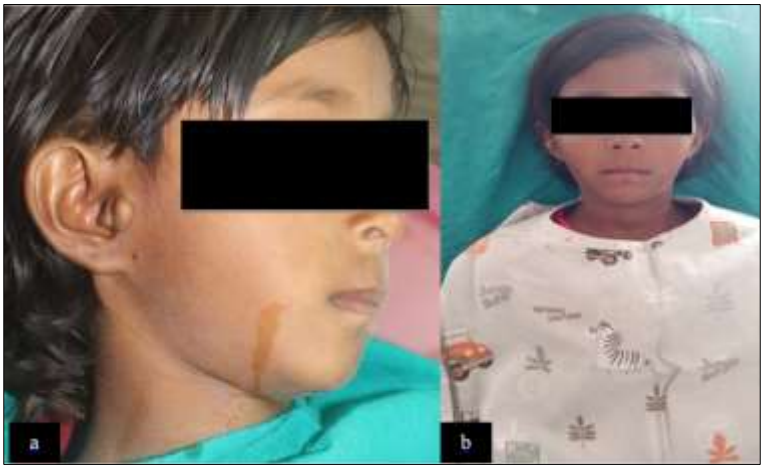

Fig 1: (a) 08-year-old female child with right accessory parotid gland with fistulous duct and pre-auricular appendage. (b) Facial asymmetry on the right side.

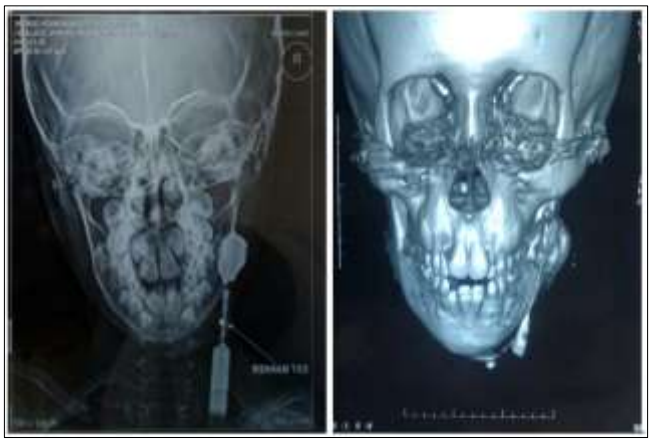

Fig 2: contrast CT sialogram showing accessory parotid gland.

Fig 3: (a) Dissection of accessory parotid duct (b) Transpositioning of accessory duct into oral cavity using Delore's method.

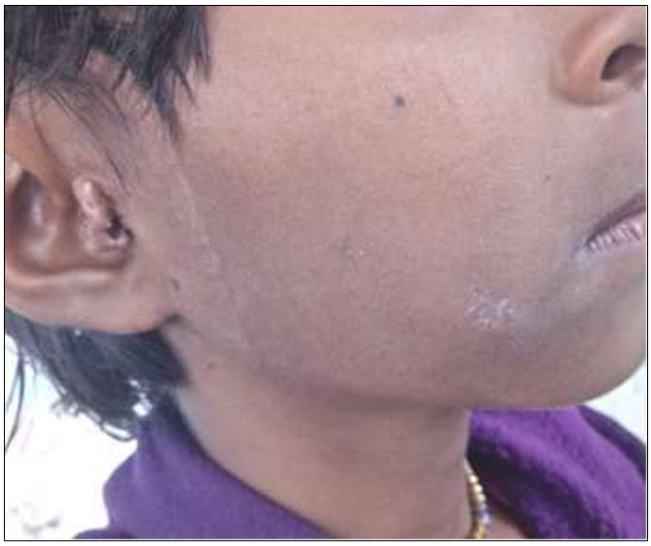

Fig 4: post-operative picture after 1 month

4. References

1. Yadav G D, Yadav A, Soni J, Chaudhary G. Accessory parotid gland with ectopic fistulous duct - Surgical view: A case report and review of current literature. Natl J Maxillofac Surg 2014;5:236-9
2. Dutta M. The ectopic accessory parotid system with congenital cheek fistula: An overview and current update. Laryngoscope. 2017; 127(6):1351-1360.
3. Currarino, Guido, & Votteler, Theodore P. (2006). Lesions of the accessory parotid gland in children. Pediatric Radiology, 36(1), 1-7.
4. Dutta, Mainak & Chatterjee, Indranil & Roy, Soham. (2018). The ectopic accessory parotid system: A new patient, studied through magnetic resonance imaging. Clinical Imaging. 48. 40-3.
5. Martelli H Jr, Miranda RT, Fernandes CM, Bonan PR, Paranaíba LM, Graner E, Coletta RD. Goldenhar syndrome: clinical features with orofacial emphasis. J Appl Oral Sci. 2010 Dec;18(6):646-9.
6. Tasse C, Böhringer S, Fischer S, Lüdecke HJ, Albrecht B, Horn D, Janecke A, Kling R, König R, Lorenz B, Majewski F, Maeyens E, Meinecke P, Mitulla B, Mohr C, Preischl M, Umstadt H, Kohlhase J, Gillessen-Kaesbach G, Wieczorek D. Oculo-auriculo-vertebral spectrum (OAVS): clinical evaluation and severity scoring of 53 patients and proposal for a new classification. Eur J Med Genet. 2005 Oct-Dec;48(4):397-411.
7. Beleza-Meireles A, Clayton-Smith J, Saraiva JM, Tassabehji M. Oculo-auriculo-vertebral spectrum: a review of the literature and genetic update. J Med Genet. 2014 Oct;51(10):635-45.
8. Dutta, M. The ectopic accessory parotid system with congenital cheek fistula: An overview and current update. The Laryngoscope, 2016, 127; 1351-360