Collaural Fistula : A Case Report

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ABSTRACT

Introduction

Collaural fistula or cervico-aural fistula is rare and accounts for less than 8% of branchial cleft anomalies. Their rarity and diverse presentations have frequently led to misdiagnosis and inappropriate treatment.

Case Report

We report one such case of a 7 year old girl who presented to us with two discharging cutaneous openings on the left side; one in the floor of the left external auditory canal and another in the upper neck and lower face (infra-auricular region).

Discussion

Surgical exploration and excision is the definitive treatment of a collaural fistula. A sinus/ fistula opening into the external auditory canal, should be removed with skin and cartilage. If more than 30% of the circumference of the external auditory canal is denuded, split thickness skin grafting and stenting are recommended. The potential post-operative complications are facial nerve paralysis and recurrence of the lesion. Fistulogram is a useful diagnostic tool.

Keywords

Branchial Region/abnormalities; Collaural Fistula

The external ear canal is a derivative of the first branchial arch. Anomalies of the first branchial cleft, therefore involve external ear structures which are normally managed in otology practice. However in duplication anomalies, clinical features are varied and patients may present to a General Surgeon or a Maxillofacial Surgeon.

Embryological anomalies of the first branchial cleft usually present as cysts, swellings or fistulas in the preauricular or postauricular area or high in the neck, which may become infected. Failure to recognize these unusual cases may result in misdiagnosis, inadequate treatment and subsequent recurrence. This paper reports a case to highlight specific diagnostic clinical features of collaural fistula with special reference to embryology and histological classification and relevant surgical management in Otorhinolaryngology.

Case Report

A 7 year old girl, presented with a discharging sinus in the left external auditory canal. (Fig. 1) But on careful examination, it was found that there were two cutaneous openings; one opening in the floor of the external auditory canal in the cartilaginous portion and another on the left upper neck/ lower face in infra-auricular (parotid) region. At presentation there was serous discharge from both the openings.

Before surgical excision, Methylene Blue injected from the cervical opening was seen coming out of the opening in the external auditory canal. Surgical excision was carried out through a horizontal skin crease incision including an ellipse surrounding the cutaneous neck opening. Dissection was done along the subcutaneous plane, with the track being carefully separated from the surrounding tissue. (Fig. 2) Methylene blue, which was injected prior to surgery served as a useful guide to trace the tract. It was found that the tract was passing immediately lateral to the trunk of the facial nerve just before it entered the parotid gland, inferior to the angle of mandible. Hence, nerve preservation was accomplished.

On tracing it higher, the fistulous tract was found to be attached to the floor of the external auditory canal. The fistula was completely excised and sent for HPE. Post-operative recovery was uneventful. Histopathological examination of the excised fistula confirmed that it was

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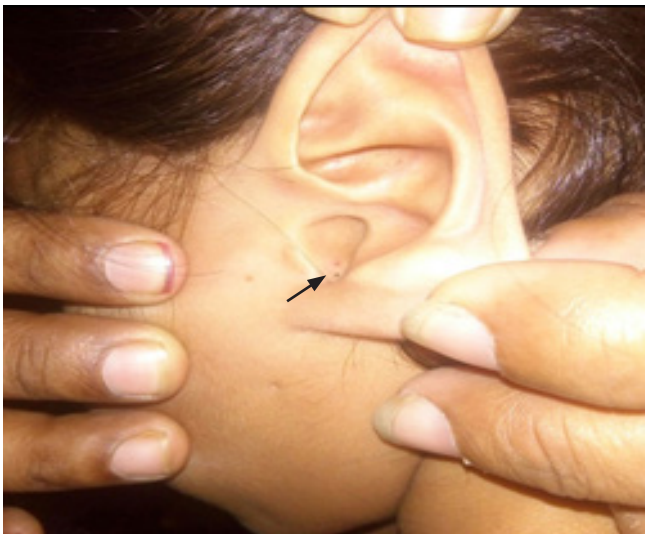

Fig.1 The opening in the floor of the external auditory canal (black arrow)

lined by squamous epithelium with adnexal structures. (Fig. 3)

Discussion

Incidence: More than 200 cases are reported in the literature. It accounts for less than 8% of all branchial abnormalities.[1,2]

Embryology: During the 4th week of human embryological development, 6 pairs of branchial arches appear which will form the future lower face and neck,[2] and they disappear by the 7th week. Mesodermal in origin, these arches are separated from each other by the 5 branchial clefts (ectoderm) externally and 5 pharyngeal pouches internally (endoderm). First branchial cleft anomalies are a result of incomplete closure of the cleft.[3] The chance of malformations occurring nearer the ear and parotid is greater than that occurring at the hyoid region, as obliteration of the cleft proceeds from ventral to dorsal.[4] Although the lesion normally has a close relationship to the parotid and facial nerve, the relationship is variable, presumably because of temporal differences during development.[3]

Classification: In 1971, an anatomic classification by Arnot[5] designated Type I anomalies as defects in the parotid region, appearing during early or middle adult life. Type II defects appear in the anterior cervical triangle with a communicating tract to the external auditory canal and usually develop during childhood.

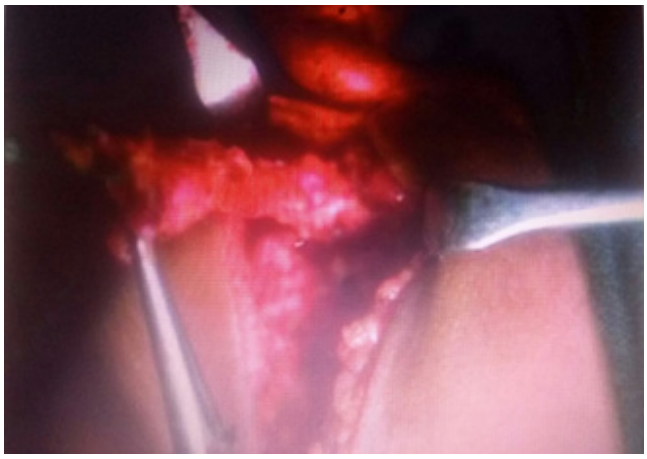

Fig.2 Sinus tract being excised

In 1972, Work proposed a histological classification.[6] Type I anomaly is a defect of ectodermal origin, arising from duplication of the membranous external auditory canal. It can have a tract running medial and parallel to the external auditory canal, superior to the facial nerve and sometimes extend to the mesotympanum. Type II defects are ectodermal and mesodermal in origin, containing skin with adnexal structures as well as cartilage. They present as cyst, sinus, fistula or a combination. They are associated with a sinus/ fistula

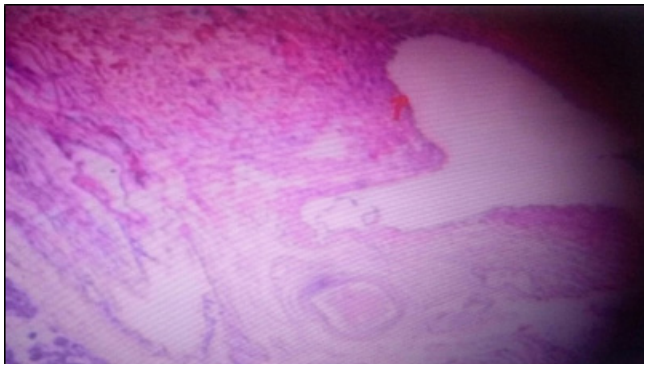

Fig.3 Histopathology slide showing the tract lined by squamous epithelium with adnexal structures (H&E, 100x)

opening in the region of the submental triangle, extend superiorly through the parotid gland towards the floor of the external auditory canal at the level of bony-cartilaginous junction or the cartilaginous portion.

Although congenital in origin, first branchial cleft anomalies can present later in life, at a mean age of 18.9 years.[1] Clinically, they may manifest as a cystic swelling or discharge from a fistulous opening either pre-auricularly, in the cheek, or post-auricularly, or high in the neck.

Otorrhoea is the most frequent otological symptom. A thorough otological examination should be performed in all cases as this may reveal a pit visible in the external canal. A sinus/fistulous opening in the external auditory canal is present in only 44% of patients, and even if such an opening exists, it may not necessarily appear obvious.[4] Sinuses and fistulae arise from incomplete closure of the first branchial groove. In 2 out of 3 cases reported by Sichel et al, first branchial cleft anomalies were associated with a myringeal web, an epidermal structure which extends from the floor of the external auditory canal to the umbo of the tympanic membrane.[7] However, in a larger series reported by Triglia et al, it was found in only 10% of the patients.[4]

For a Type II lesion, an early identification of the facial nerve at the stylomastoid foramen is recommended. If this part is affected by a disease, identifying the facial nerve proximally in the temporal bone and tracing it distally are probably the safest option. The relationship of the lesion to the facial nerve is variable. In a series of 10 patients with first branchial cleft anomalies reported by Solares et al, 7 lesions ran medial to the facial nerve, 2 were lateral and 1 ran in between the branches of the facial nerve.[3] Fistula has a tendency to run deep into the nerve, whereas sinus tracts tend to run laterally to it. Due to its variable relationship with the nerve, its removal warrants an early identification and a wide exposure of the nerve,[4] and/or the use of facial nerve monitoring.

Accurate diagnosis of first branchial cleft cysts located in and around the parotid gland can be difficult without surgical exploration.[4] Poncet's triangle is the anatomical triangle where first branchial cleft cysts or sinus orifice are typically located.[7] The limits of the Poncet's triangle are the external auditory canal above, the mental region anteriorly and the hyoid bone inferiorly.

Should the sinus/ fistula opening involve the external auditory canal, it is removed with skin and cartilage. A primary closure is normally possible but if more than 30% of the circumference of the external auditory canal is denuded, split thickness skin grafting and stenting are recommended. If tympanic membrane or middle ear structures are involved, a reconstructive otologic surgery may be necessary.

Surgical exploration and excision is the definitive treatment of these defects.[6] The potential post-operative complications are facial nerve paralysis and recurrence of the lesion. Fistulogram is a useful diagnostic tool. Imaging studies especially CT Scan and MRI is useful for showing the tract near to the facial nerve.[7]

Conclusion

Cysts, sinuses or fistulous tracts which are pre-auricular or post-auricular or high in the neck, may represent a branchial cleft anomaly. An inflammatory process in the region of Poncet's triangle should immediately raise an index of suspicion. The usefulness of various anatomical and histological classifications is limited and it is often difficult and confusing trying to correlate the clinical picture with various classifications. The conventional operation for first branchial cleft fistula is highly complicated and facial nerve palsy is a frequent complication.

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