

CASE REPORT

First branchial cleft cyst – The misdiagnosed anomaly

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INTRODUCTION

Branchial apparatus consists of paired branchial arches separated by branchial clefts externally and branchial pouches internally.^[1,2] Persistence of cleft and pouch both results in a fistula with an internal and external opening. Cysts, swellings, or fistulas in the upper part of the neck may be the presentations of first branchial cleft anomalies, which may become infected hence delayed or misdiagnosed, leading to complications during surgery.^[3] Branchial cleft anomalies are divided into two groups based on the presence or absence of mesothelial elements within the wall. Lesions of ectodermal origin, present medial to the concha, are classified as Type 1 while Type 2 lesions are of both ectodermal and mesodermal origin and contain cartilage and hair follicles. The lower opening in Type 2 lesion is usually below the angle of mandible.^[4]

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ABSTRACT

First branchial cleft anomalies are the rare defects which account <8% of the branchial anomalies. There are varied presentation of these anomalies in the form of cystic swelling, discharging sinuses, and fistulas in the preauricular and cervical region. Due to its varied clinical manifestations and rarity, misdiagnosis and delayed diagnosis are common leading to incomplete and inappropriate management. We are presenting a case of a 6-month-old child with branchial cleft anomaly with delay in definitive treatment due to misdiagnosis.

KEY WORDS: Branchial apparatus, branchial anomaly, branchial cleft cyst

CASE REPORT

A 6-month-old female child was referred to us with complaints of incessant crying and discharge from the left ear since birth, which was intermittent, profuse, occasionally foul smelling, associated with redness, and excoriation of the left ear. There was no history of trauma to the ear. On examination, the left ear appeared to be macerated with purulent, foul-smelling discharge coming from EAC [Figure 1]. There was a small tag in post-auricular area. There was no external opening in the neck. Tympanic membrane was normal. Due to common occurrence of acute otitis media in this age, the child was advised conservative treatment multiple times by local medical practitioner with little symptomatic improvement. Computed tomography scan was suggestive of a cystic lesion present in the left post-aural and infra-auricular region [Figures 2 and 3]. On the basis of clinical as well as radiological investigation, diagnosis of first branchial cleft anomaly was made.

After complete pre-anesthetic investigations and checkup, the patient underwent surgical excision of the cystic lesion under general anesthesia. On tracing it higher, the cyst was found to be getting attached to the floor of the external auditory canal

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near the lobule. The cystic lesion with sinus tract was excised in toto [Figures 4-6]. Post-operative period was uneventful. There was mild facial weakness which was recovered completely. Follow-up after 1 month was excellent [Figure 7].

DISCUSSION

First branchial cleft anomalies are rare comprising <5% of all branchial cleft anomalies.^[1] It develops due to incomplete obliteration of cleft between the mandibular processes of the first and second arch. Type I lesions are due to duplication of cartilaginous



Figure 1: Macerated left external auditory canal with a small post-aural tag



Figure 4: Macerated external auditory canal and pinna



Figure 2: Computed tomography scan coronal section showing cystic lesion in infra-auricular area

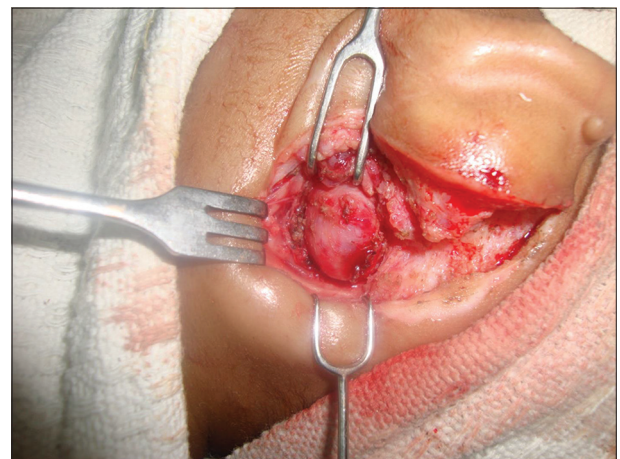


Figure 5: Intraoperative visualization of cystic lesion

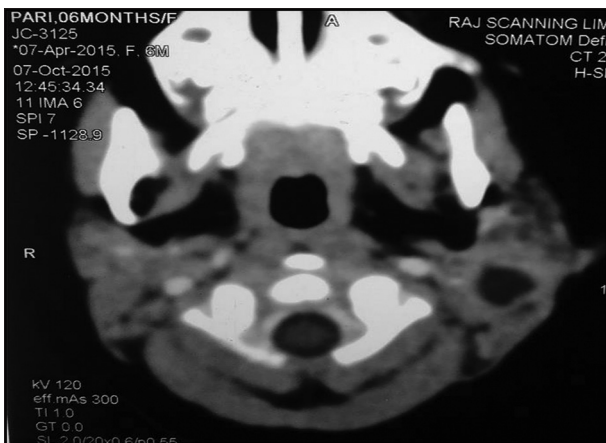


Figure 3: Computed tomography scan axial section showing cystic lesion

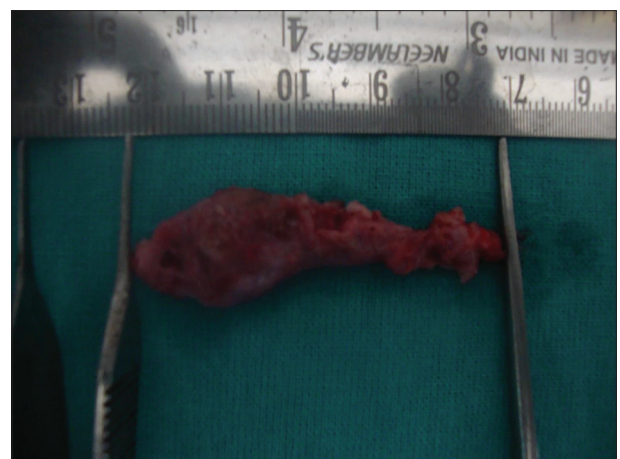


Figure 6: Excised specimen



Figure 7: Post-operative picture after 1 month

external auditory canal. A cystic mass in the post-auricular area extends anteromedially along the external auditory meatus, passes lateral to the facial nerve, and ends at the bony meatus. Sometimes, there is no external opening except after infection. Type II lesion is considered to be a duplication of the cartilaginous external auditory meatus and pinna. In this lesion, sinus tract passes from an external opening in the neck along the anterior border of SCM muscle, superficial, or deep to the facial nerve. It can open in to the external auditory canal, which is called the collaural fistula. Although these lesions are congenital, they may present later in life. Both types of anomalies may present as recurrent mass or as a draining sinus. They may present with repeated episodes of infection, which, in turn, may present itself as cystic swelling or discharge from a fistulous opening. A pre-operative sinogram/fistulogram is advisable for confirmation of position of the upper end. The patient may present as otorrhea or consequences such as maceration of pinna as in our case. There might be a history of repeatedly incision and drainage of an apparent abscess in upper neck.

Facial nerve identification is very important in cases of type 2 lesions.^[5] Surgical excision of the tract in toto is the treatment of choice, ideally before acute infection or abscess has supervened. The upper end of the fistulous tract has an unpredictable relationship to the external auditory canal cartilage and may terminate at the canal or form a duplicated canal parallel and inferior to the external auditory canal proper. In case, the sinus/fistula opening involves the external auditory canal, it is removed with skin and cartilage.

CONCLUSION

Abnormal development of the branchial apparatus can lead to the formation of different anomalies, which may be asymptomatic or may present later in life. Diagnosis is rather easy with a proper knowledge of the anatomy of the branchial anomalies but sometimes it is delayed due to varied presentations. Strong suspicion is very important to diagnose such congenital anomalies because of their various presentations. Extent of the tract should be properly known before surgery because of the vital structures of the neck in relation to these lesions. Surgery is the treatment of choice as these lesions have a high incidence of recurrent infection and further complications.

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