Case Report

First branchial cleft fistula in a 7 years old girl

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First branchial cleft anomalies are rare, occurring in less than 8% of all branchialabnormalities. Usually, these anomalies diagnosed in early adulthood period with variable presentations like cervical drainage from a pit-like depression at the angle of the mandible, parotid enlargement due to inflammation and auricular signs like recurrent otitis externa and ear discharge.Wereport a 7 yearsold female patient who was found to have first branchial cleft fistula, presenting with left ear discharge and mucoid discharge from a tiny opening below the angle of the left mandible. (Rawal Med J 201;41:274-275).

Keywords: First branch ialfisula, discharge, branchial cyst.

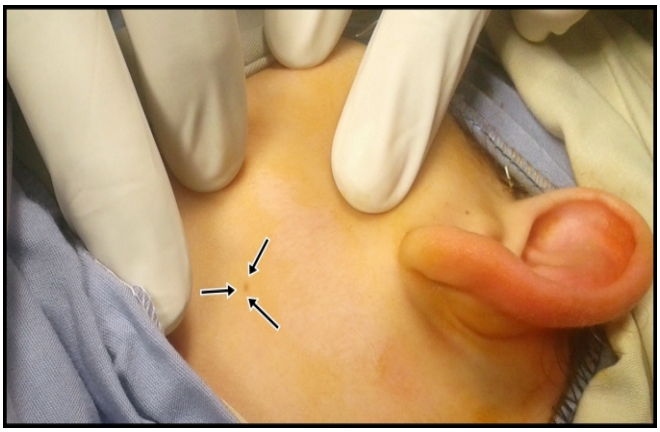
INTRODUCTION

First branchial cleft congenital anomalies are rare, with less than 8%of branchialabnormalities.[1-3] Overall incidence is less than one per million.[4] First branchial cleft anomalies are common in females (69%) than males (31%) and fistulas occur more frequently on the left side (64%).[5,6] The first branchial arch forms the mandible and the maxillary process of the upper jaw while the first branchial cleft forms the tympanic cavity, middle ear cavity, and the mastoid air cells.[1,2] Presentation can include cervical signs like drainage from a pit-like depression at the angle of the mandible, parotid signs resulting from rapid enlargement due to inflammation and auricular signs like recurrent otitis externa and ear discharge.[6,7] The tract may be passing through the parotid gland close to the facial nerve branches which make resection difficult.[6] Repeated surgery places the facial nerve at greater riskof iatrogenic injury due to scars and fibrosis, emphasizing the importance of complete resection at the first attempt.[5-7]

CASE PRESENTATION

A healthy 7 years old girl presented to pediatric surgery clinicwith history of left ear discharge and intermittent mucoid drainage from a tiny opening below the angle of the left mandiblesince few weeks.Physical examination revealed 1x1mm pit just below the angle of the left mandible (Fig. 1). Left ear examination revealed intact tympanic membrane with a small opening in the floor of the external auditory canal just lateral to the bony-cartilage junction.High resolution temporal CT scan was inconclusive. She was planned for intra-operative fistulogram and complete resection of the fistulous tract. The parentswere informed about the details of the operation, the possibility of facial nerve damage and superficial parotidectomy.

Fig. 1. A pit just below the angle of mandible.



The externalostium of the fistulous tract was circumcised, fistula tract was reached and was canulated. Dissection was kept close to the fistula tract to avoid injury to the parotid gland and facial nerve branches. There was no need to perform an intra-operative fistulogram because the anatomy was very clear. The internal ostium was excised by

pediatric otology surgeon.

Post operative recovery was uneventful with no signs of damage to the branches of facial nerve.The fistulous tacthistopathological examination confirmed the diagnosis of branchial cleft fistula.She was followed in the clinic one month, 6 months and one year later andshe was completely asymptomatic.

DISCUSSION

Baer was the first who describe branchial anomalies in 1827.[4] Till now there is a controversy about the cause and etiology of branchial cleft anomalies,[8] but in general, it is due to incomplete fusion of ventral portion of first and second arches.[9] Arnot classified branchial anomalies anatomically into two types; type 1 in parotid area which present in adulthood and type 2 in cervical triangle and external auditory canal, which present in child period.[3]Other classification system consider tissue type lining the tract: Type I contains ectoderm, type II has both ectoderm and mesoderm.[1,6]

Lesions may present as a fistula, a sinus, or a cyst. Fistulas occur slightly more commonly (52%) than sinuses (48%).[5,6]Despite these, in congenital anomalies, the diagnosis is late with mean age of 19 years.[4,8] So our case is considered a special one because we diagnose it very early (7 years).Less than 200 cases were reported in literature, most of them were diagnosed in adulthood,[9]and usually adult head and neck surgeons deal with these cases, but in this case,the patient was treated by a pediatric surgeon with excellent results.

Computed tomography (CT) is the best modality for the diagnosis of these anomalies and give us more details about the extension of tract and boney involvement.[4]Surgical excision of the tract and, in some cases, resection of a small amount of skin and cartilage within the external auditory canal are the best treatment and usually result in a total cure.[5,7] Recurrence occurs if the tract is not excised completely.[7] So, it is critical for the surgeon to understand the extension of the lesion and the course of fistulas or sinus tracts and related structures.[5-7]

In summary,first branchial anomalies are uncommon but we must consider it for correct diagnosis and management due to vital structures related to it.Pediatric surgeons can deal with these anomalies perfectly with excellent results.

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Conflict of Interest: None declared
Rec. Date: Oct 9, 2015 Accept Date: Jan 11, 2016

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