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OTOLOGY

FIRST BRANCHIAL CLEFT ANOMALY-PRESENTING AS RECURRENT POSTAURICULAR SWELLING

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ABSTRACT First branchial cleft anomalies encompass 10% of all branchial cleft defects. Due to misdiagnosis, management is often inadequate, recurrence is common and iatrogenic injuries of the facial nerve occur. A 26 year old female presented with recurrent, painful, postauricular swelling with right sided facial nerve palsy. Incision and drainage was performed twice in this region previously. A solitary, firm, tender swelling of 5x2cm in size, was seen in the right postauricular region with a sinus opening superior to the swelling. Investigations included HRCT temporal bone. She underwent excision of the cyst and fistulous tract. Histopathology report showed features consistent with epidermoid keratinous cyst. They should be suspected in patients with a cutaneous opening defined by the hyoid bone inferiorly, the sternocleidomastoid muscle posteriorly and the mandible anteriorly. Early diagnosis is needed to avoid recurrent infection and secondary development of a fistulous tract. Permanent cure requires complete surgical excision

KEYWORDS : first branchial cleft anomaly, post-auricular swelling.

INTRODUCTION

First branchial cleft anomalies are a relatively uncommon group of congenital malformations of the head and neck, accounting for between 5% and 12% of all branchial cleft defects.[1–3] Many theories have been proposed to describe the abnormal embryogenesis that results in their formation but the branchial theory is the most popular today.[4,5] The branchial apparatus is a transient structure that is present during weeks four to seven of foetal development which contributes to the formation of many head and neck structures. It consists of six mesodermal branchial arches that are separated externally by branchial clefts or grooves and internally by pharyngeal pouches, all lined with ectoderm. The first branchial cleft is the only one to contribute to structures in the adult, and it persists as the epithelium of the external auditory canal. It is thought that anomalies result from incomplete obliteration of this cleft and the degree of closure governs whether the lesion takes the form of a cyst, sinus, or fistula.[5,6] We report on a case of first branchial cleft anomaly, which displayed some of the complexities of this condition.

CASE REPORT

A 26 year old lady, presented with history of swelling in the right post auricular region. The swelling was associated with pus discharge and pain. She gave history of two similar episodes in the past four years for which incision and drainage was performed. On examination, a solitary, soft to firm, fluctuant, tender swelling of 5x2cm in size, was seen in the right postauricular region with a sinus opening found superior to the swelling. Pus discharge was seen extruding from the sinus opening(fig.1)

Fig 1: Pre-operative image of the post-auricular region

Fine needle aspiration was done from the swelling, which revealed keratin debris. An HRCT of the temporal bone was taken, which showed a fistulous tract between the sinus opening and the external auditory canal. The scan also suggested significant erosion of the bone over the sigmoid sinus. After explaining the operative procedure including its prognosis and taking relevant consent the patient was prepared for excision of the fistulous tract under general anaesthesia. Methylene blue dye was injected into the sinus opening so as to identify the entire tract. Intra operatively, a well encapsulated and lobulated cyst was dissected, extending and connecting to the floor of the external auditory canal with extrusion of foul smelling discharge (fig.2). The sigmoid sinus was found to be anteriorly placed and eroded (fig.3). She had an uneventful post-operative period. Post-operative histopathological examination findings were consistent with that of an epidermoid keratinous cyst.

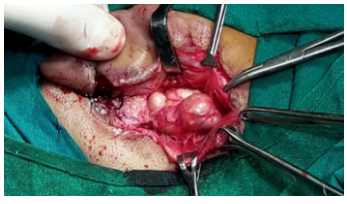

Fig 2: Intra-operative image depicting the cyst after dissection

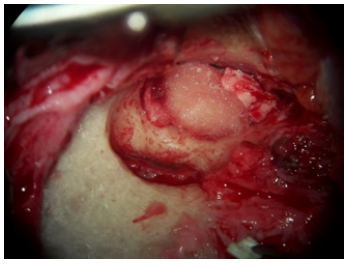

Fig 3: Anteriorly placed and eroded sigmoid sinus

DISCUSSION:

First Branchial cleft anomalies are classified into two types. Type I lesions are cystic (ectodermal origin) and are lined by squamous epithelium with or without accessory skin structures.[7] They are considered to be a duplication anomaly of the membranous external auditory canal. Lesions are usually associated with the parotid gland and lie superficial to the facial nerve.[8] Type II anomalies (ectodermal-skin and mesodermal -cartilage origin) present as a sinus, cyst or fistula and are considered to be a duplication of the membranous and cartilaginous external auditory canal.[8] Lesions extend from the submandibular region, over the ramus of the mandible, through the parotid gland with variable relation to the facial nerve terminating below the skin of the ear canal as a sinus.[9,10] these anatomical relations ordain clinical presentation and surgical management.

Unfortunately, first branchial cleft anomalies are often misdiagnosed, with majority of patients suffering from a long clinical course before diagnosis and definitive surgical management. Most patients present between birth and the second decade with swelling in the periauricular (24%), parotid (35%), or cervical regions (41%).[10] There may be extrusion of pus or desquamating epithelial debris, or a skin pit noted in the case of a sinus or fistula.[11] There is often a history of recurrent infection that required a course of antibiotics and repeated incision and drainage. Various authors have reported a delay of 11 months and 4 years between initial presentation and management.[2,6,10] 35–48% of cases undergo repeated surgical procedures with a mean of around 2.5 operations being done before a cure was achieved.[2,10] Otological examination may show a depression, pit, or mass in the external auditory canal with history of recurrent otorrhoea, otitis externa, or hearing loss.[10,11]

It has been suggested that CT is useful to define both the location and extent of the lesion.[12-14] However, post-operative scarring may make interpretation difficult. Once the acute infection has subsided , definitive treatment is only by complete excision. Any sinus, fistula, or areas with skin breakdown should be excised in continuity. Sometimes, a small portion of the skin and cartilage of the external auditory meatus is also removed to prevent recurrence.[12-14]

The chance of secondary infection of branchial lesions is about 25% and is usually recurrent.[15] This poses a problem when excision is finally performed as dissection becomes difficult due to post-operative scarring and adhesions. Extensive adhesions between the lesion and facial nerve can lead to facial palsy postoperatively.[6,10] The overall recurrence rate after excision for first branchial cysts is higher than that of second cleft anomalies and increased by a further 20% with history of previous infection.[15]

CONCLUSION

First branchial cleft anomalies are rare and difficult to diagnose. Early diagnosis and surgical management is imperative in case of first branchial cleft anomalies. Mismanagement by repeated drainage should be avoided as this leads to extensive scarring and adhesions that may cause facial palsy. Definitive treatment by complete dissection and excision with identification and preservation of the facial nerve must be performed

REFERENCES

1. Olsen KD, Maragos NE, Weiland LH. First branchial cleft anomalies. Laryngoscope 1980; 90: 423–436.
2. Ford GR, Balakrishnan A, Evans JNG, Bailey CM. Branchial cleft and pouch anomalies. J Laryngol Otol 1992; 106: 137–143.
3. Doi O, Hutson JM, Myers NA, McKelvie PA. Branchial remnants: a review of 58 cases. J Pediatr Surg 1988; 23: 789–792.
4. Maran AGD, Buchanan DR. Branchial cysts, sinuses and fistulae. Clin Otolaryngol 1978; 3: 77–92.
5. Benson MT, Dalen K, Mancuso AA, Kerr HH, Cacciarelli AA, Mafee MF. Congenital anomalies of the branchial apparatus: embryology and pathologic anatomy. Radiographics 1992; 12: 943–960.
6. Ikarashi F, Nakano Y, Nonomura N, Kawana M, Okura T. Clinical features of first branchial cleft anomalies. Am J Otolaryngol 1996; 17: 233–239.
7. Arnot RS. Defects of the first branchial cleft. S Afr J Surg 1971; 9: 93–98.
8. Work WP. Newer concepts of first branchial cleft defects. Laryngoscope 1972; 82: 1581–1593.
9. Belenky WM, Medina JE. First branchial cleft anomalies. Laryngoscope 1980; 90: 28–39.
10. Triglia JM, Nicollas R, Ducroz V, Koltai PJ, Garabedian EN. First branchial cleft anomalies: a study of 39 cases and a review of the literature. Arch Otolaryngol Head Neck Surg 1998; 124: 291–295.
11. Nofsinger YC, Tom LWC, LaRossa D, Wetmore RF, Handler SD. Periauricular cysts and sinuses. Laryngoscope 1997; 107: 883–887.
12. Choi SS, Zalzal GH. Branchial anomalies: a review of 52 cases. Laryngoscope 1995; 105: 909–913.
13. de Gaudemar I, Elmaleh M, Cortez A, Peuchmaur M, Francois M. Unusual presentation of a first branchial cleft. Eur Arch Otorhinolaryngol 1995; 252: 57–60.
14. Takimoto T, lwawaki J, Ishikawa S. First branchial cleft anomaly: a report of case. Auris Nasus Larynx 1989; 16: 151–155.
15. Liston SL. The relationship of the facial nerve and first branchial cleft anomalies – embryologic considerations. Laryngoscope 1982; 92: 1308–1310.