CASE REPORT

TYPE FOUR BRANCHIAL FISTULA: DIAGNOSIS AND MANAGEMENT: A RARE CASE REPORT

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ABSTRACT: Branchial cleft fistulae are rare congenital abnormalities that arise from the abnormal persistence of branchial apparatus remnants. Fourth branchial cleft fistulae pass deep to second arch structures and over third arch structures, in a direction extending from the anterior border of sternocleidomastoid (SCM) muscle to the upper pole of the ipsilateral pyriform fossa. Because of this anatomical route, these long tubular structures are intimately associated with major neuro-vascular structures in the neck. We present the clinical presentation and surgical management of a fourth branchial cleft fistula, in a 23-year-old male. Preoperative CECT fistulogram of the neck clearly demonstrated the fistula. Patient was managed using combined stepladder transcervical and intra oral fistullectomy.

KEYWORDS: Branchial fistula, branchial cyst.

INTRODUCTION: The branchial cyst, fistula, and sinuses are the anomalies of the branchial apparatus which consists of five mesodermal arches separated by invaginations of the ectoderm called as clefts. The branchial fistula is not a true fistula as it rarely has two openings. More often even if both ends are patent there is a thin membrane covering the internal opening.¹ Demonstration of a complete branchial fistula on imaging studies is uncommon.²

We describe the case of a young 23 years aged man who came to us with on and off discharge from left sided lower neck initially with subsequent development of neck swelling. His condition was initially treated with medical therapy. We were able to diagnose the branchial fistula by CT fistulogram of the opening. This case highlights the importance of making a full visual inspection of the involved sinus in order to avoid missing an unexpected, albeit a rarely encountered, pathology.

CASE REPORT: A 23-year man came to the otolaryngology department with eleven year history of, off and on discharge from left side of lower third of neck, which was associated with on and off neck swelling. There was no history of trauma and patient got operated for the same problem about 3 years back. Local examination revealed a pinhead opening along the anterior border of sternocleidomastoid on the left-hand side of lower third of neck. There was no limitation of neck movements. He was afebrile. Further systemic examination was unremarkable and his Hb14.1 gm%, total count of 6300 cells/cumm, platelet count 121000 cells/cumm, ESR 21 mm/hr, serum creatinine 0.7 mg/dl, blood urea 29 mg/dl was HIV, HBsAg negative.

There were sign of inflammation around the opening and discharge was present. Patient was put on antibiotics for a period of seven days. A CECT sinogram was done to delineate the exact course of tract before surgery. CECT sinogram revealed a fistulous tract noted from skin in lower neck anterior to sternocleidomastoid traversing anterior to carotid artery and internal jugular vein and open internally into apex of left pyriform fossa-s/o fourth arch branchial fistula.

CASE REPORT

Based on imaging and the clinical history a diagnosis of branchial fistula of fourth branchial arch was made. The patient underwent excision of the tract by a step ladder approach along with intraoral approach.

DISCUSSION: The branchial apparatus were first described by Von Baer while its anomalies were first described by von Ascheroni.³ The branchial fistula is an uncommon anomaly of embryonic development of branchial apparatus. Amongst these, anomalies of second branchial arch as well as pouch are common. They represent 90–95% of branchial anomalies.⁴ During embryonic development, the second arch grows caudally and it covers the second, third, and fourth branchial clefts. The cervical sinus of his is formed by the fusion of this second arch with the enlarging epi pericardial ridge of the fifth arch. The edges of cervical sinus in the due course fuse and hence in life no defect is seen. However, it is the persistence of intervening ectoderm that gives rise to branchial cyst. The branchial fistula results from break down of the endoderm, usually in the second pouch. In the normal course a persistent fistula of the second, third and fourth branchial cleft and pouch passes from the external opening in the mid or lower third of neck in the line of the anterior border of the sternocleidomastoid muscle, deep to platysma along the carotid sheath.

The tract then passes medially deep between the internal and external carotid arteries, after crossing over the glossopharyngeal and hypoglossal nerves. Finally, it opens internally in the tonsillar fossa in case of second pharyngeal pouch, into larynx in case of third pharyngeal pouch, into pyriform fossa in case of fourth pharyngeal pouch. Clinically most of these cases present with mucoid discharge from the anterior aspect of neck. In addition presence of infection may lead to formation of an abscess and signs of inflammation at the site of opening. Although branchial fistula may occur at any age but commonly they present in first or second decade of life.⁵ Most of the times it is a simple sinus opening that extend up the neck for a variable distance. Complete branchial fistula with internal opening into pyriform region is rare. Besides radiological demonstration of complete tract is further difficult. Ford et al⁴ documented one case in their series of 98 fistulae in which the entire tract was visualized.

The diagnosis is most often clinical and radiological investigations are done to confirm the course of tract. However, a fistulogram if performed delineates the tract and it is often the commonest investigation available as done in our case. A complete fistula demonstrable by a fistulogram is uncommon.² With the available of multislice computed tomography scan a CT fistulogram with reformatted images unambiguously delineates the relation of sinus tract to that of important structures of neck. It also helps in classifying the type of lesion, provides a road map for surgeon prior to surgery, and reduces the chance of recurrence. MRI is most advantageous for Type I first branchial cleft cysts and for parapharyngeal masses that may be second branchial cleft cysts. By the inherent nature of better tissue contrast it provides the relationship of glandular tissue to the mass (e.g. fat planes between the parotid gland and a parapharyngeal mass) and hence acts as a roadmap prior to surgery. Ultrasound is useful in situations where CT scanning and MRI are unavailable. Ultrasound can delineate the branchial cleft cyst.

However, complete delineation of fistulous tract is difficult even with the use of high resolution transducers. Although ultrasound can confirm the cystic nature of a mass, it does not adequately evaluate the extent and depth of neck lesions.⁶ The treatment of choice for branchial fistula is surgical excision.

CASE REPORT

Several surgical approaches have been described for the management of a branchial fistula. These include a transcervical approach, either by a stepladder approach or through a long incision along the anterior border of sternocleidomastoid and a combined pull through technique. The higher incision should be bigger than the lower one because the fistula tract is deeper in location in the vicinity of important neurovascular structures. The reported incidence of recurrence rate was 3% where only external approach was used. This most probably is due to incomplete excision of the fistula tract in the parapharyngeal space.⁴ However, no recurrence rate is reported after using combined oral and transcervical approach.

Our patient's clinical picture was discharging sinus at left lower cervical region with no regional lymphadenopathy which was later on confirmed on CT FISTULOGRAM as type 4 branchial fistula. CT fistulogram revealed a fistulous tract noted from skin in lower neck anterior to sternocleidomastoid traversing anterior to carotid artery and internal jugular vein and open internally into apex of left pyriform fossa-s/o fourth arch branchial fistula. Patient was managed through combined approach using stepladder transcervical and pull through intraoral fistullectomy.

CONCLUSION: Branchial fistula arising from fourth branchial arch is rare. Diagnosis of patient was made through clinical presentation and CECT sinogram which revealed type 4 branchial fistula. Patient was managed using combined stepladder transcervical and intraoral fistullectomy.

REFERENCES:

1. Shekhar C, Kumar R, Kumar R, Mishra SK, Roy M, Bhavana K. The complete branchial fistula: a case report. *Ind J Otolaryngol Head Neck Sur* 2005; 57: 320.
2. Augustine AJ, Pai KR, Govindarajan R. Clinics in diagnostic imaging. *Sing Med J* 2001; 42: 494–495.
3. De PR, Mikhail T. A combined approach excision of branchial fistula. *J Laryngol Otol* 1995; 109: 999–1000.
4. Ford GR, Balakrishnan A, Evans JN, et al. Branchial cleft and pouch anomalies. *J Laryngol Otol* 1992; 106: 137–143.
5. Ang AH, Pang KP, Tan LK. Complete branchial fistula. Case report and review of the literature. *Ann Otol Rhinol Laryngol* 2001; 110: 1077–1079.
6. Ahuja A, King AD, Metriweli C. Second branchial cleft cyst: variability of sonographic appearances in adult cases. *Am J Neuroradiol* 2000; 21: 315–319.

CASE REPORT

Pre-operative

CT- Fistulogram

Intra operative

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