A rare bilateral second arch branchial anomaly

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Abstract

Branchial cysts, sinuses, and fistulas are rare anomalies, and bilateral presentation is even more uncommon, consisting of only two to three percent of all cases. There have been several reports of bilateral branchial cysts or sinuses, but only a few cases of bilateral branchial fistulas have been reported. But in our review of literature, we were not able to come across a similar case of a branchial fistula on one side and a branchial sinus on the other. The most comparable occurrence we could find was of a simultaneous second branchial cleft fistula on the left and right-sided sinus with cystic dilatation.

Keywords: Branchial, Cyst, Fistula, Sinus

Introduction

Branchial arch anomalies are typically seen in the pediatric age group, although patients may present these at any time throughout their adulthood. We report a case of a bilateral branchial anomaly of concurrent second arch branchial fistula and sinus.

Case Report

A 29-year-old man presented with a swelling on the right side of the neck, and discharge from bilateral openings in the lower neck (Figure 1) since childhood. On examination, the swelling was located in the lower one-third of the neck on the right side. The swelling was round, measured about 1 centimeter in diameter, soft, non-tender, and non-fluctuant. The discharge was insidious in onset, intermittent, scanty, watery in nature, non-blood stained but since the past six months the patient was having yellowish discharge from the opening on the right side. There was no history of recurrent upper respiratory tract infection, fever, difficulty in swallowing, or hoarseness of voice.

Ultrasound revealed evidence of an anechoic tubular tract measuring 3 mm in cross-section seen along the anterior border of the left lower sternocleidomastoid muscle. It extended from the superior border of the thyroid cartilage, and terminated inferiorly near the insertion of sternocleidomastoid. On the right side, ultrasound showed evidence of an elongated cystic lesion measuring 6.8 cm in length, and 1 cm in cross-section. It was seen along the anterior border of the

Figure 1: Bilateral external opening in the lower neck

How to cite this article: Suresh P, Ashish C A, S G Mahesh, Shaharyar A M. A rare bilateral second arch branchial anomaly. MJMS. 2017; 2(1): 29-31.
right lower sternocleidomastoid muscle extending from the superior border of the thyroid cartilage, to the insertion of sternocleidomastoid inferiorly. The major vessels of the neck were normal. There was no evidence of calcification, ductal dilation or focal lesions.

A course of antibiotics was prescribed, on completion of which, the yellowish discharge from the right side subsided. On the basis of clinical findings, the case was originally diagnosed as congenital bilateral second arch branchial sinuses. A bilateral tract excision was performed under general anesthesia. During the procedure, methylene blue was injected into the external openings of the branchial cleft tracts to determine the extent and to explore the possibility of internal openings (Figure 2). Under direct laryngoscopy, an internal opening was identified over the post-pillar of the tonsil on the left side (Figure 3), but there was no evidence of an opening on the right side, concluding the final diagnosis of a branchial fistula on the left, and a branchial sinus on the right. The histopathology report of the excised specimens revealed the following: specimen from the left side showed a tract lined by granulation tissue and a cystic space lined by respiratory epithelium. Lymphoid tissue with follicle formation was evident in the wall. Specimen from the right side showed a fistulous tract of granulation tissue connected to a collapsed cyst, which was lined by respiratory epithelium showing squamous metaplasia. Adjacent lymphoid follicles with reactive centers were seen.

**Discussion**

Branchial cysts, sinuses, and fistulas are uncommon embryonic developmental anomalies, which are seldom seen in clinical practice. Rarely, two to three percent of cases have bilateral presentation. Of the various anomalies, branchial cysts are most frequently noted. Cysts are typically seen in males after the age of 10 years peaking in the third decade of life and often present unilaterally, on the left side. In contrast, branchial sinuses or fistulas most frequently present in neonatal age group or at an early age. Unlike cysts, fistulas occur more commonly on the right side and are more prevalent in women. The occurrence of fistulas is the rarest of the branchial anomalies. Complete fistulae are uncommon as in the majority of cases the tracts end blindly. Since they have only one opening, they are correctly termed as branchial sinuses. A complete second arch branchial fistula is one that has an external opening and a definite internal opening in the tonsillar area. Lesions of the second branchial arch are commonly present as a lateral neck swelling, or a discharging sinus that may be superimposed by an infection; located between the upper two-thirds and lower one-third of sternocleidomastoid. In the case of a sinus or fistula, there is a fistulous tract which extends upward in the deep tissues of the neck for variable distances from the site of the external opening. The typical tract of second branchial anomalies runs deep to the platysma muscle between the second and third arch structures; ascending along...
the carotid sheath and passing between the internal and external carotid arteries, and crossing above the glossopharyngeal and hypoglossal nerve and over the stylopharyngeus muscle. A cystic dilatation lined with stratified squamous or columnar epithelium, and occasional granulation tissue or ectopic salivary tissue may lie anywhere along the pathway. The tract may end or exit in close proximity to the tonsillar fossa, although it is rare to see a tract with an internal opening in the tonsillar region, as was seen in our case on the left side. Accordingly, the tract is labeled as a cyst, sinus or fistula. It is important to note that the tract of a fistula may be blocked by thick secretions or granulation tissue, making it difficult to identify a fistula which maybe misdiagnosed as a sinus. It is helpful to pass a guide wire through the fistulous tract, to determine the extent of it, in case an internal opening is not easily identified followed with dye injection or fluoroscopic imaging.

Conclusion

Only few cases of bilateral branchial fistulas have been reported in the English medical literature. Though there have been several reports of bilateral branchial cysts or sinuses, in our review of literature we were not able to come across a case similar to our presentation. The most comparable occurrence we could find was of a second branchial cleft cyst on the left, and fistula with cystic dilatation on the right. As far as we know, our case of concurrent second arch branchial fistula on one side and sinus on the other is the first of its kind to be reported. With proper surgical planning and technique, a good result can be achieved.

References