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Case Report Open Access

Bilateral Branchial Apparatus Anomaly – A Rare Case

Hegde SS*, Keshav Prasad and Kulamarva Rama Sandeep

Kasturba Medical College, Mangalore-575003, India

Abstract

The Branchial apparatus has been well described and held accountable for many congenital anomalies of the head and neck region. A congenital fistula is not as commonly encountered as a cyst or sinus, and a complete congenital fistula from the third branchial apparatus is a rare occurrence. According to the branchial apparatus theory, the course of a branchial fistula is derived from its embryological origin. Here, we report a case of a complete branchial fistula of the right side and a branchial sinus on the left side, which is a rare occurrence.

Case Report

A 20-year-old man presented with a history of discharge from right and left sides of lower part of his neck (Figure 1) since birth. The patient had bilateral neck swelling over the same site at the time of birth, which burst open spontaneously on the 12th day of his life. Since then the patient has on and off discharge from the aforementioned sites. The discharge increases after food intake. The discharge is whitish, viscous, non-foul smelling, painless and non-blood stained. The patient had consulted a local physician for the above complaints and has received antibiotic course multiple times and he was advised to undergo surgical treatment for the same. The treatment was not sought due to financial constraints. There was no history of fever, recurrent upper respiratory tract infections, dental or jaw infections. There was no history suggestive of Tuberculosis or Malignancy. General Physical Examination revealed a level 5 lymph node measuring 1 cm × 1.5 cm, which is soft, tender and mobile. Vitals and systemic examination were within normal limits. Local examination revealed a sinus on right side measuring 1 cm × 2 cm located between the two heads of the right sternocleidomastoid muscle. On the left side the sinus measured 0.1 cm × 0.2 cm. No active discharge from the sinuses at the time of examination. It moved with deglutition and protrusion of tongue. Oral cavity examination was within normal limits. Routine blood investigations and chest X-ray were within normal limits. The patient was taken to the operating theatre for exploration and excision of the fistula. Under general anesthesia, the patient was put in Rose's position. The external opening of the tract was dilated with a probe to delineate the tract. Probing was done with a number 5 infant feeding tube on both sides following a circular incision (Figure 2), which identified a right sided Branchial fistula and a Branchial sinus on left side. On the right side, dissection was done by deepening the incision and platysma was explored. Dissection extended upto the angle of mandible. Fistulous tract was identified lying superficial to right sternocleidomastoid, facial vein, greater auricular nerves and ansa-cervicalis (Figure 3). Fistula was cut and ligated at the upper end of dissection, probing was done with a guide wire. Tract was extending upto the right tonsillar bed which was identified by laryngoscopy by applying intermittent traction and visualizing the tonsils. Excision of the fistula was done (Figure 5). A sub-platysmal drain was placed and the sub-cutaneous tissue, platysmal and skin were approximated with 3-0 Ethilon. A circular incision applied over the lower left side of the neck. The tract was explored by blunt dissection and was found to be extending into the sub-platysmal plane 4cms below the left clavicle (Figure 4). The sinus was cut beyond its blunt end and the tract was excised (Figure 5). Wound was closed in layers and the skin sutured with 3-0 Ethilon. The patient was extubated, movement of the vocal cords were bilaterally equal. The patient was shifted to post-operative ward in a stable condition. The patient was then put on parenteral antibiotics for 5 days. The post-operative period was uneventful and the patient recovered completely. The patient was discharged on the 15th post-operative day. A Histopathological examination of the specimen confirmed the presence of a tract lined by squamous epithelium.

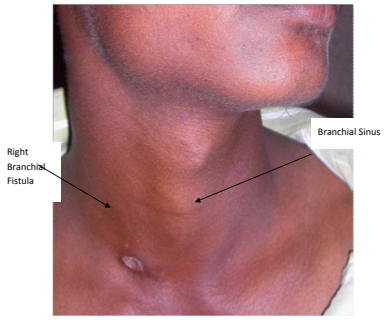

Figure 1: A complete branchial fistula of the right side and a branchial sinus on the left side.

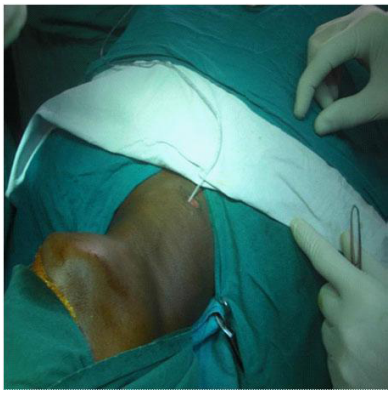

Figure 2: On table insertion of Infant feeding tube into the right branchial fistula.

***Corresponding author:** Shruti S Hegde, A-506, RNS Shanti Nivas, Tumkur Road, Post Yeshwanthpur, Bangalore-560022, India, Tel: 09483465685; E-mail: raregem15@gmail.com

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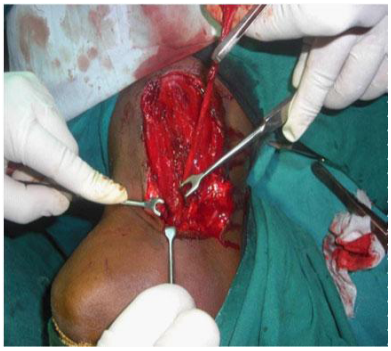

Figure 3: Dissection of Right side Branchial Fistula.

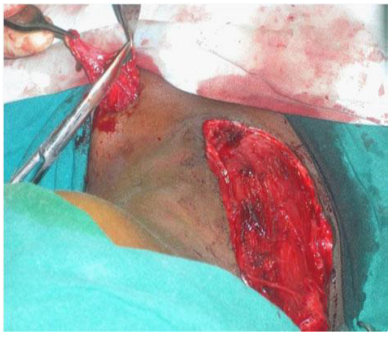

Figure 4: Dissection of Left side Branchial Sinus.

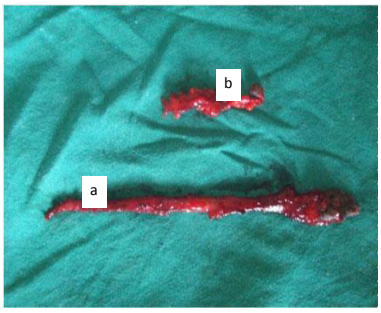

Figure 5: Dissected right branchial fistula (a) and left branchial sinus (b).

Discussion

The Branchial apparatus comprises six arches with the mesoderm as its core, separated by clefts and pouches on the ectodermal and endodermal sides, respectively [1]. Maldevelopment of the branchial apparatus leads to branchial anomalies that occur in the form of a sinus, cyst or fistula. These anomalies may originate from the first to fourth cleft/pouch, with the commonest (95%) arising from the second cleft/pouch [2]. Our patient had a true fistula on right side with both internal and external openings and a sinus on left side. He had a history of neck swelling during infancy, which was similarly documented by other authors as a first presentation in their cases [3]. He later presented with a discharging fistula during childhood [3]. A Branchial fistula is thought to form when the mesenchyme that separates the cleft and pouch involutes, thus uniting them [4]. Therefore, the fistula would be caudal to the structures derived from the corresponding arch and dorsal to the structures from the following arch. Branchial fistulae are rare in clinical practice. They present with mucopurulent discharge from an opening between the upper two-thirds and lower one-third of sternocleidomastoid [5] and can recede back into the neck on swallowing [6]. They are more common on the right side and more prevalent in women [7]. Complete fistulae are uncommon as in the majority of cases the tracts end blindly [7]. These are termed Branchial sinuses. Most authors recommend either an ultrasonography of the neck, barium swallow or CT imaging for a branchial anomaly. Some authors even advocate the use of magnetic resonance (MR) imaging [8-10]. No imaging modalities could be done prior the surgery in this case, as the patient cannot afford it. The treatment of choice for a third branchial fistula is surgical resection, as there has been no evidence of spontaneous regression and this condition is at a high risk of recurrent infection. The standard method of excision is the use of a stepladder incision. Another method is the stripping of the tract. Stripping of the branchial fistula involves passing the stripper inside the tract using wire stilletes, vein strippers or arterial intimal strippers. Its advantage lies in that it is a simple method and does not involve the use of extensive dissection [11]. However, it is a blind technique that poses a higher risk of injury to the surrounding structures, especially with a branchial fistula that had recurrent infection with surrounding fibrosis. Regardless of the technique used, complete removal of the tract is essential to avoid recurrence. The tract may be delineated intraoperatively using methylene blue dye injection. Some authors, however, find this technique suboptimal due to the extravasation of dye into the surrounding tissue [12,13]. Another method of delineating the tract is by using the Fogarty catheter; however, maintaining a catheter in the tract during surgery is difficult. Edmonds et al. [13] have recommended the use of direct laryngoscopy which was used in our case. The recurrence rate of Branchial anomaly is 3% for a primary lesion and as high as 22% for lesions with previous infection and surgery [14]. Recurrence arises mostly when the thyroid tissue is not removed or when the tract is not identified [4]. Another complication of surgery is injury to the recurrent laryngeal nerve. Our patient had an uneventful post-operative period with complete recovery and no recurrence in first year follow-up period.

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