

Bilateral Second Branchial Arch Fistula in a 19 year old - A Case Report

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SUMMARY

Developmental anomalies involving the branchial apparatus result in branchial cysts, sinuses and fistulas. A congenital branchial fistula is not as commonly encountered as a cyst or sinus. Branchial fistulas mostly arise from the second branchial arch and present at birth. Only few cases are bilateral. Complete branchial fistulas with both external and internal openings are rare. We present a case of bilateral branchial fistula with both external and internal openings in a 19 year old male which is a rare occurrence.

KEYWORDS: Branchial anomalies, bilateral branchial fistula, adult.

Introduction

Branchial apparatus develops during the 2nd - 6th week of gestation. It consists of 6 mesenchymal arches separated by ectodermal clefts externally and endodermal pouches internally. Each branchial arch, cleft or pouch will develop into a specific structure within the head and neck region. Incomplete obliteration of the branchial arches, clefts and pouches leads to development of branchial anomalies which presents as cysts, sinuses or fistulas. A branchial cyst is formed from epithelial rest, it contain retained secretions and has no internal or external opening. A branchial sinus results from a persistent cleft (external sinus) or a persistent pouch

(internal sinus). Persistence of both cleft and pouch will result in a fistula.

Branchial anomalies may originate from the first to the fourth branchial cleft/pouch. Fistulas of the second branchial cleft are the most common branchial anomalies, accounting for up to 90% of all branchial cleft fistulas, with only 2%-3% being bilateral. Complete branchial fistula with external and internal openings is rare, more often even if both ends are patent there is a thin membrane covering the internal opening. The external opening lies along the middle and lower third of anterior border of sternocleidomastoid muscle. The tract then ascends along the carotid sheath between the external and internal carotid arteries behind the posterior belly of digastric muscle and anterior to hypoglossal nerve and terminates in the pharynx as a slit on the anterior part of cephalic half of the posterior faucial pillar. Although branchial fistulas may occur in any age group, commonly patients present in the first or second decades of life. Sixty percent are on the left and 40% are on the right, although bilateral branchial fistulae have also been described.

The symptoms associated with these tracts include persistent or intermittent mucoid drainages, recurrent infection leading to

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cellulitis and abscess formation. Ultrasound, computed tomography (CT) with or without contrast, fistulogram or magnetic resonance imaging (MRI) may aid in the diagnosis⁴. Recently, multidetector CT fistulography has been described for diagnosis of branchial cleft fistula.

Attempt at treating the fistula was made in the past by using sclerosants, this was associated with increased risk of perforation and damage to surrounding structures.

Total surgical excision of the fistula is the treatment of choice, which often requires meticulous dissection with multiple incisions (stepladder pattern) combined (cervical and intraoral pull through) with endoscope assisted surgery. Endoscopic cauterization is a minimally invasive technique to obliterate the internal opening of the fistula, it can be done as a single modality or in combination with open surgery^{4,8,9}.

The cauterization is done using diathermy probe, low power diode laser, or chemicals e.g. trichloroacetic acid or silver nitrate. Complications of the surgery include recurrence (3 % in fresh cases to up to 20 % in second surgical attempt), injury to facial, hypoglossal, glossopharyngeal and spinal accessory nerves, injury to internal jugular vein, carotid vessels and hematoma formation.

We present a rare case of bilateral branchial fistula in a young adult male.

Case Presentation

A 19 year old man presented with a history of bilateral pinpoint opening at the left and right aspects of the lower neck since birth. He has associated recurrent whitish mucoid discharge from the openings. There was recurrent swelling on the left which started 10 years ago with associated pain, fever and odynophagia which usually rupture

discharging pus. He had several course of antibiotics with some relief but symptoms kept recurring. There was no family history of similar illness. Examination revealed a small opening in the anterior border of the sternocleidomastoid muscle on the right at the junction of its upper 2/3 and lower 1/3 discharging mucoid material. There was an abscess on the left at about the same level with the opening on the right that was tender with skin changes over it and containing pus (fig.1). An assessment of bilateral branchial fistula complicated by abscess formation on the left was made. The abscess was drained and patient treated with antibiotics and anti-inflammatory drugs. The patient was then prepared for excision under general anaesthesia after the abscess has healed on three weeks conservative management. Methylene blue dye was injected into the tracts after cannulation to aid tract identification during dissection. An elliptical incision was made around the two openings with a horizontal skin crease incision connecting the two elliptical incisions. The tracts were then followed upward using blunt and sharp dissection (fig.2). On the left it was followed up to the tonsillar fossa, a left tonsillectomy was done and the tract delivered through the tonsillar fossa (fig. 3). While on the right it was followed to its superior limit and excised.

The left fistula tract measured 12cm while the right measured 10cm in length (fig. 4 and 5 respectively). The mucosal opening in the oropharynx was closed with vicryl 3/0 and neck wound closed in two layers after inserting a vacuum drain. Post-operative period was uneventful. The histology of the specimen revealed pseudostratified columnar epithelial and squamous metaplasia of the epithelium lining of the branchial cyst with no evidence of malignancy (Fig. 6&7).

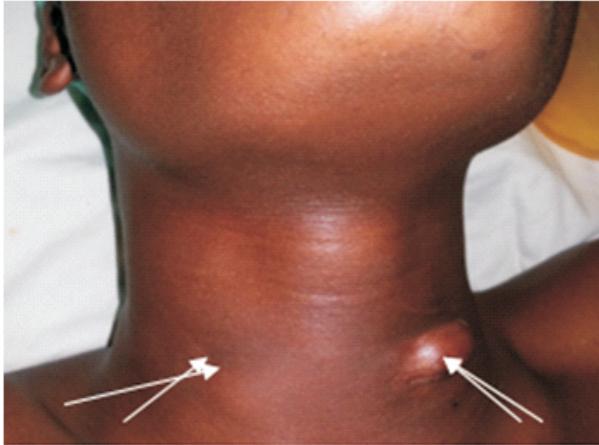


Fig. 1: Picture showing the sinus opening on both sides



Fig. 4: Right fistula tract

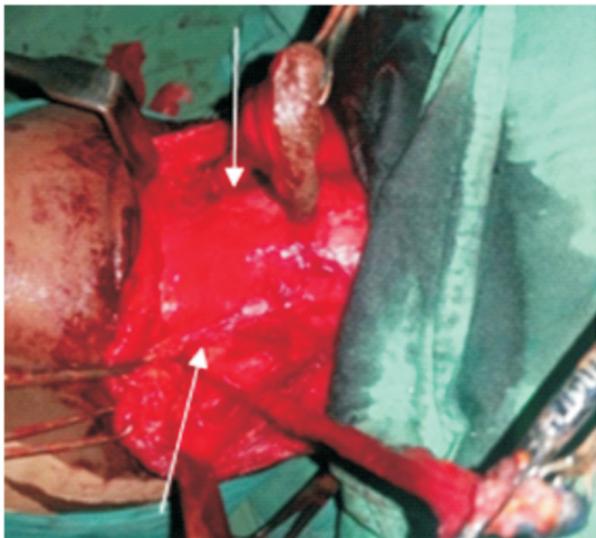


Fig. 2 : Both tracts dissected superiorly indicated by the arrows.

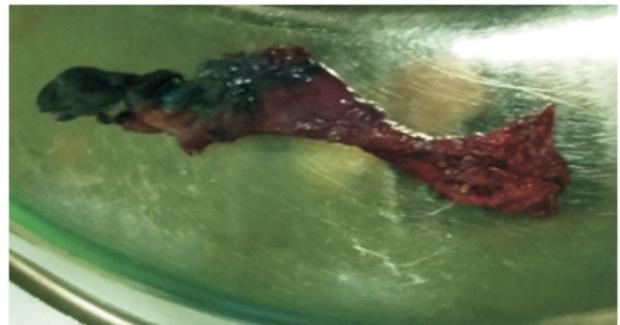


Fig. 5: Left fistula tract

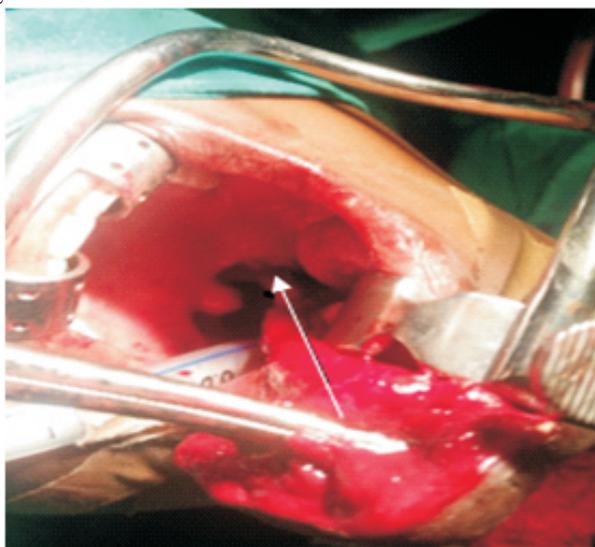


Fig. 3: Tract delivered through the tonsillar fossa

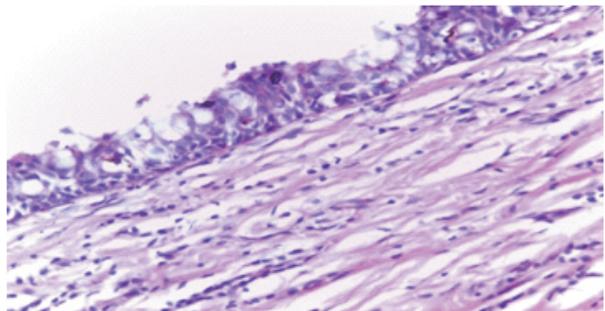


Fig. 6: Photomicrograph showing Pseudostratified columnar epithelium lining branchial fistula on the right. H&EX400

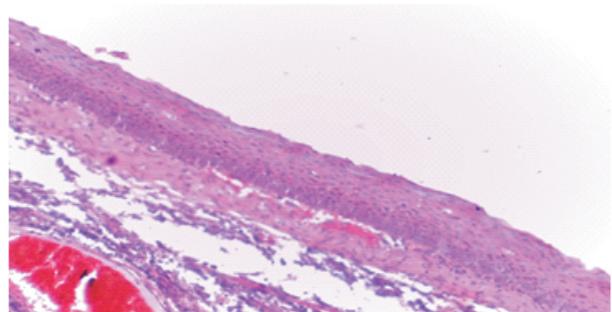


Fig. 7: Photomicrograph showing Squamous metaplasia epithelium lining of branchial fistula on the left. H&EX200

Discussion

Branchial apparatus consist of 6 mesenchymal arches with mesodermal core separated by ectodermal cleft externally and by endodermal pouches internally. Mal-development of these arches result in branchial cleft anomalies, which presents as cysts, sinuses or fistulas. Fistulas of the second branchial cleft are the most common branchial anomalies, accounting for up to 90% of all branchial cleft fistulas, most are unilateral with only 2%–3% being bilateral making them rare. Branchial cleft fistulas mainly presents as a fistulous tract on the skin of the neck anterior to the sternocleidomastoid as in this case, typically at its middle third.⁶ Discharges from opening on both sides of the neck as seen in this patient, was also reported.³ In addition our patient had recurrent abscess formation on the left side with associated fever, pain and odynophagia that require drainage and antibiotics prior to surgical excision. A similar presentation of recurrent discharge associated with fever has been documented¹². This patient had bilateral second cleft fistula with no family history of neck cyst or fistula, which was similarly reported. In contrast most reported bilateral cases are familial. Thus this may be an isolated case. Surgical excision is the treatment of choice as there has been no report of spontaneous resolution and the condition

is associated with recurrent infection, cellulitis and abscess formation.⁶ Excision through a wide cervicotomy incision remains the method of choice for treatment of second branchial cleft fistulas.¹¹ In our case, a horizontal skin crease incisions connecting the two elliptical incisions around the fistula openings, this gave us adequate exposure of the tracts and structures on both sides, which facilitates accurate dissection without the need for stepladder incisions or other incisions on both sides. The tracts were traced to the posterior part of the tonsillar fossa on the right but fell short of that in the right. Complete tract excision was achieved on the left and high tract ligation was done on the right. Left tonsillectomy was done to allow complete excision and delivery of the tract through the oral cavity, this was similarly reported. This allowed complete excision of the whole tract with less chance of recurrence. In contrast complete excision of the fistula has been reported without the need for tonsillectomy.^{4,7} We recommend tonsillectomy if the tract can be traced to the tonsillar fossa to avoid leaving an inner tract lead to recurrence. Our case is a rare variety with fistula occurring bilaterally requiring meticulous dissection and tonsillectomy for complete removal. By this method recurrence is reduced significantly.

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