International Journal of Surgery Case Reports 97 (2022) 107365

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

A complete second branchial fistula in a four years old child

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ARTICLE INFO

Keywords:
Branchial apparatus
Complete fistula
Step ladder
Case report

ABSTRACT

Introduction and importance: Second branchial anomalies either cyst, sinus, or fistula are the top differential diagnosis of lateral neck masses or swelling in pediatrics age group. Yet, it is very rare for the branchial fistula to have two openings.

Case presentation: Here we present a four years old child diagnosed with complete branchial fistula by CT scan with dye injection throughout the fistula tract. We successfully managed him by complete surgical resection. Also, we provide the current literature that aids in the diagnosis and treatment of complete second branchial fistula.

Conclusion: Complete second branchial fistula is not that common anomaly; however, we must consider it as a differential diagnosis in any lateral neck masses. Complete surgical resection, step ladder approach, which will minimise the recurrence rate is the treatment of choice.

1. Introduction

Von Baer was the first one to describe the branchial apparatus, but von Ascheron was the one who describe the anomalies in its development [1]. Branchial apparatus consists of the outer most layer which called clefts and it's covered with ectoderm; the first cleft will become an epithelial layer covering the external auditory canal. Medial to it there is mesodermal core called arches; those arches will develop into varies structures such as connective tissue, muscles, bones and certain cranial nerves. Lastly, the pouches are the most inner layer of the branchial apparatus, it is covered by endodermal tissue, which will give way to lining epithelium and glands [2–4].

During embryogenesis, the second arch grows caudally to envelop the third, fourth, and sixth arches; to form the cervical sinus by fusing with the ectoderm caudal to these arches. The ectoderm inside the fused tube disappears, while the edges of cervical sinus fuse. If the ectoderm inside the tube did not disappear this will result in branchial cyst. And if the endoderm breakdown and communicate with skin or the mucosa this will results in branchial fistula [3,5].

Most of the branchial apparatus anomalies arise from the second branchial cleft [6–8]. However, complete second arch fistula with internal opening into the tonsillar region is rare [7,8].

Here we present a case of complete second branchial fistula which extend from right anterolateral aspect of the neck till the right tonsillar pillar. Patient presented to ENT outpatient clinic, Al-Farwaniya hospital, which is a community hospital located in Al-Farwaniya, Kuwait.

This work has been reported in line with the SCARE 2020 criteria [9].

2. Case report

A four years old Kuwaiti boy, previously healthy present to ENT outpatient department, Al-Farwaniya hospital, Kuwait complaining of thick discharge from an opening in the right side of the neck. This opening was noticed by his mother at birth. The discharge was on and off from the opening, especially after upper respiratory tract infection. The mother also noticed recurrent swelling in the right side of the neck. The swelling was painless, progressively increasing in size. No skin change was noticed in the skin surrounding the opening. No history of trauma. This was the first time he sought medical care regarding this complaint.

- Past medical history: no past medical history.
- Past surgical history: no past surgical history.
- Drug history: not on any medications.
- Family history: no family history of similar issue.
- Immunized up to date.

At the time of examination, pt. was conscious, oriented, and alert. A small, pinhead opening along the anterior border of sternocleidomastoid muscle on the lower 3rd of the neck, just 3 cm above medial end of the

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https://doi.org/10.1016/j.ijscr.2022.107365
Received 14 May 2022; Received in revised form 25 June 2022; Accepted 25 June 2022
Available online 28 June 2022

right clavicle was noticed (Fig. 1). There was neither active discharge nor swelling, redness, tenderness, or skin changes.

Our differential diagnosis was branchial anomalies in the first place, either cyst, sinus, or fistula. Or lymphatic malformation.

Patient was admitted, renal function test was done before requesting compound tomography imaging with contrast.

Creatinine was within the normal range. CT scan with contrast done and reported as a suggestive of a second branchial fistula (Fig. 2).

He was booked for right branchial fistula resection with tonsillectomy under general anesthesia, step ladder approach. The surgery done by ENT consultants, at ENT department, Al-Farwaniya hospital (Figs. 3–10).

Histopathology reported as right branchial cleft fistula.

3. Follow up and outcomes

Patient was discharged on Augmentin 457 ml syrup 5 ml twice daily for 7 days and adol syrup 5 ml three time per day for 7 days and was asked for follow up after one week post operation after that monthly follow up. Patient was improving without any complications.

4. Discussion

Branchial apparatus consists of three layers: the outer most layer lined by ectoderm; clefts and the inner most layer lined by endoderm; pouches, between them there is a mesodermal lining layer called arches. This was first described by Von Baer [10]. Anomalies involving branchial apparatus are not fully understood and not clear. There are many theories regarding the origin and classification of these anomalies, yet the most accepted one is the incomplete involution of branchial apparatus during embryogenesis [11].

Branchial anomalies count for 20 % of pediatrics congenital neck masses. It may present as cyst which is a remnant of ectoderm and epithelial lining space with no opening into neither the skin nor the mucosa, or it may present as a sinus which is a blind tract into the skin or the mucosa, or fistula a communication between a persistent pouch and cleft to develop [2,11,12].

Second branchial anomalies are the commonest, which cover around

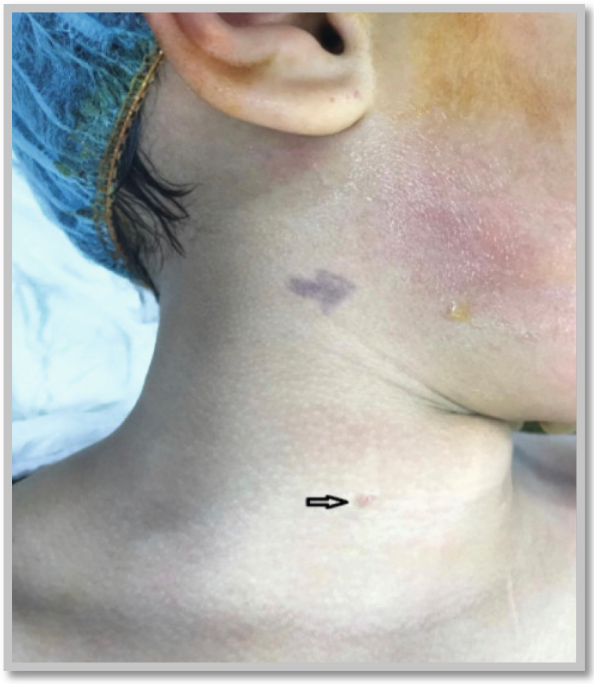

Fig. 1. An opening noticed in the right aspect of the neck.

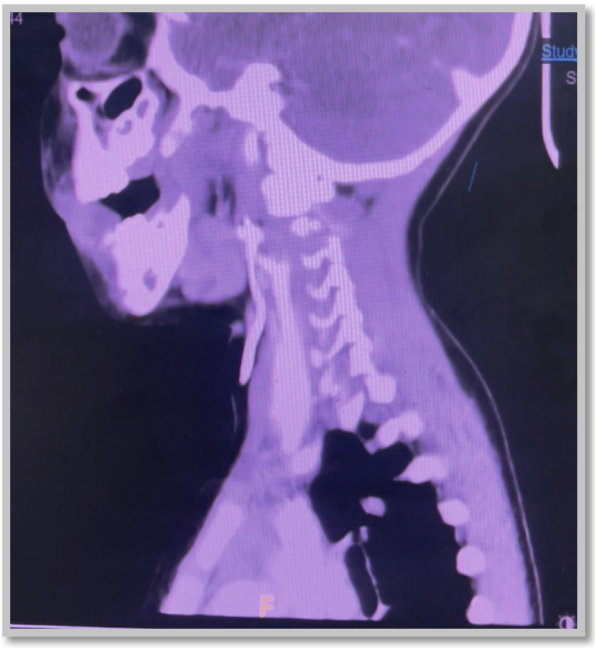

Fig. 2. Evidence of an elongated tube approximately 5.0 cm long, well defined tube extending from the right anterolateral aspect of the neck (anteromedial to the right sternocleidomastoid, anterior the carotid vessels, and lateral to thyrohyoid cartilage). Cranially terminated posterior to the right tonsillar pillar.

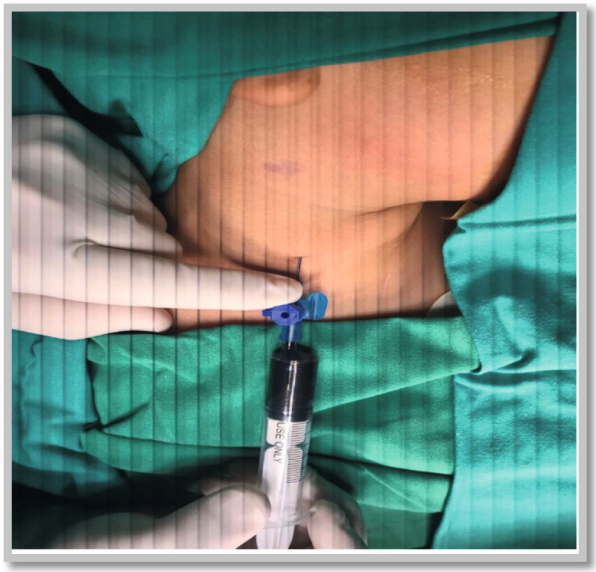

Fig. 3. Injection with methylene blue through the fistula tract was done.

95 % of all branchial anomalies [2,3,6]. A second branchial anomalies is classified into four subtypes according to Bayley's classification.

- Type 1: Anterior to the sternocleidomastoid muscle and deep to the platysma.
- Type 2: Deep to the sternocleidomastoid muscle and lateral to the carotid sheath (the most common).
- Type 3: Lies between the internal and external carotid arteries up to the lateral pharyngeal wall.
- Type 4: Medial to the carotid sheath. Lying in the pharyngeal mucosal space.

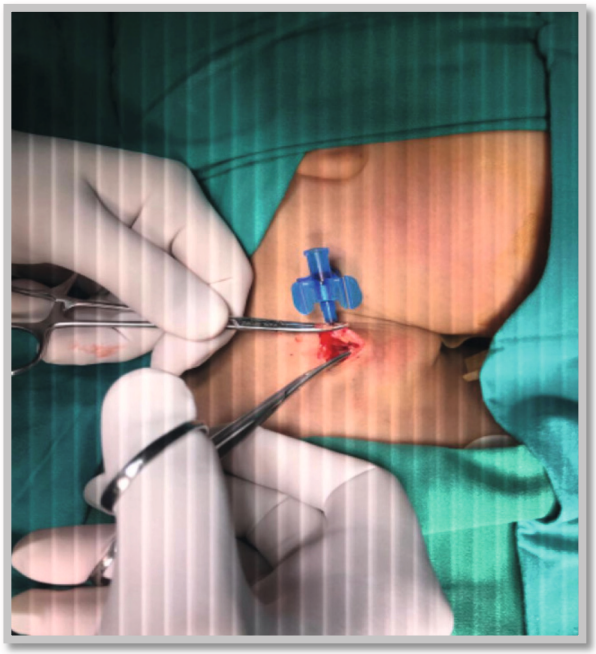

Fig. 4. Incision done.

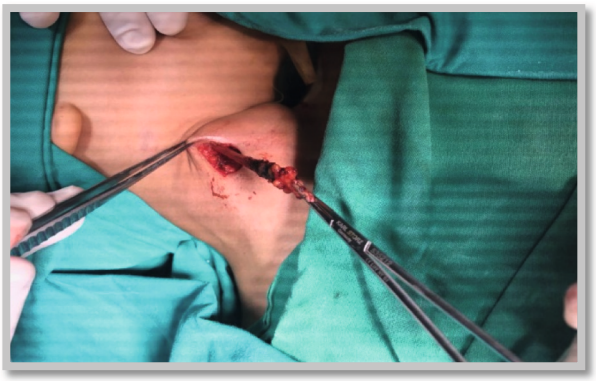

Fig. 5. Dissection.

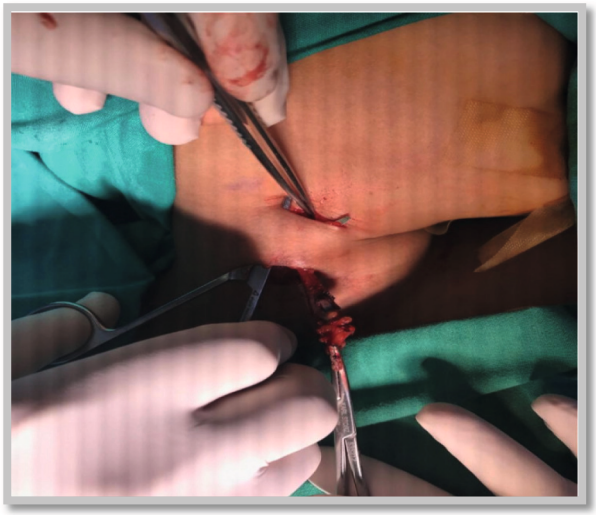

Fig. 6. Step ladder or 2nd incision.

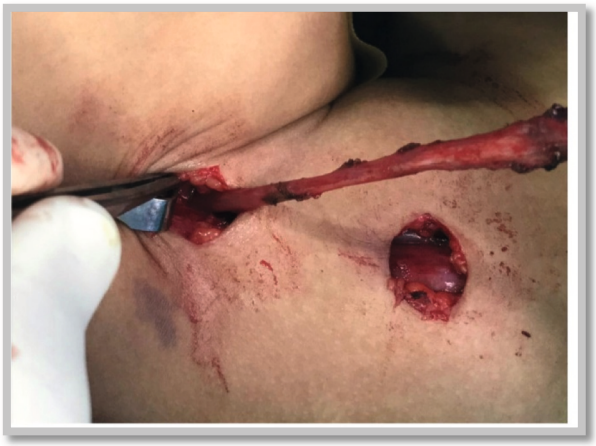

Fig. 7. Dissection.

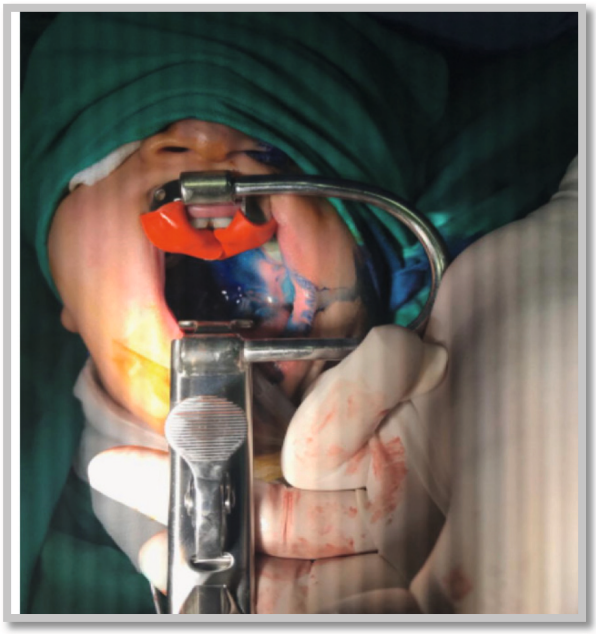

Fig. 8. Preparation for intraoral dissection with mouth gag.

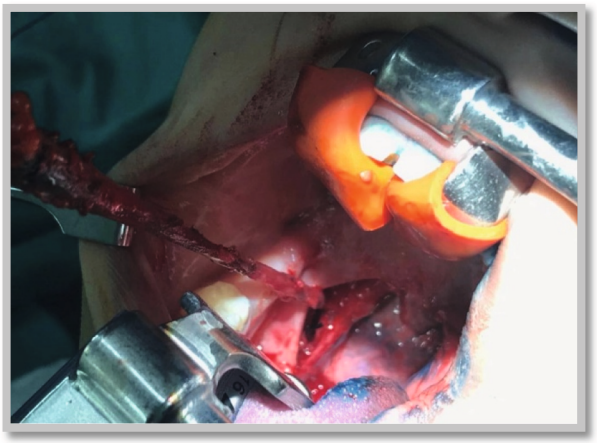

Fig. 9. Intraoral dissection.

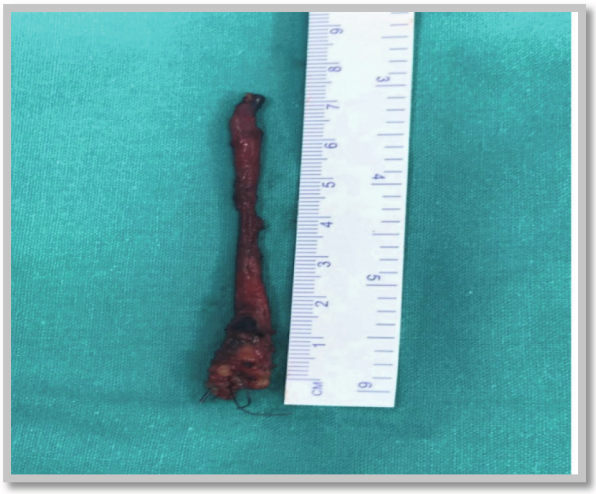

Fig. 10. Excision of the fistula tract.

However complete fistula that extended from the tonsil into the anteromedial surface sternocleidomastoid is rare [1,5,13].

Proper history taking, imaging studies, and histopathology are the best ways to diagnose second branchial fistula.

Affected patients commonly present within the first two decades of life. Most of the fistulae are clinically apparent before the age of 5 years [14]. Patients present with persistent or intermittent mucoid or mucopurulent discharge from an opening in the lateral aspect of the neck following an upper respiratory tract infection mostly [5,15,16]. Visualization of the fistula tract can be done through injecting a dye or fistulogram. Sometimes muscle relaxation [17] or stagnation of secretion due to inflammation which will obstruct the fistula [16] might give us a false negative study. CT scan with either an oily or water-soluble contrast medium injection through the fistula tract must be done prior to surgery to demonstrate the tract, and the anatomical relationship between the tract and varies important surrounding structure [18,19].

Several surgical approaches have been described for the management of a branchial fistula. However, the stander surgery for a second branchial arch fistula is the stepladder approach. It is done through two incisions in the neck that gives exposure of the fistula tract with less tissue dissection. The higher incision must be bigger than the lower to avoid the damage to important neurovascular structures [13,15,19,20].

5. Conclusion

In conclusion, second branchial anomalies are the most common cause of lateral neck masses in children. However, complete fistula is rare. An opening in lateral aspect of the neck that drains mucopurulent discharge is the commonest presenting symptom. Worldwide, the gold standard diagnostic study is CT scan with methylene blue injection through the fistula opening. And the gold standard way of management is complete surgical resection, step ladder approach, which will minimise the recurrence rate.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

Not declared.

Funding

This work did not receive any grant from funding agencies in the public, commercial, or not-for-profit stories.

Guarantor

Bashayer Alsaeedi.

Research registration number

Not applicable.

CRediT authorship contribution statement

All authors contributed toward data analysis, drafting, and revising the paper, gave final approval of the version to be published, and agree to be accountable for all aspects of the work.

Declaration of competing interest

There are no conflicts of interest to declare by any of the authors of this study.

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