Endoscopic management of fourth branchial arch anomaly: A case report

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1. Introduction

The branchial apparatus of a developing fetus, comprising six paired arches, clefts, and pouches appears by the fourth week of gestation. The mesoderm of the arches separates the ectoderm-lined clefts externally and endoderm-lined pouches within the embryo.¹

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Abstract  Fourth branchial cleft anomaly is a rare condition usually presented as recurrent neck abscess and suppurative thyroiditis. A variety of treatment options were reported in the literature. Here we present a case of a 15 year old female with left sided fourth branchial cleft anomaly. She was unsuccessfully managed by frequent surgical procedures with 6 incidences of recurrence in 2 years before applying the proper management described in this paper.

This paper reports a rare case of fourth branchial cleft anomaly with description of presentation and diagnosis and demonstration of the effectiveness of endoscopic management with a long follow up duration (23 months) with no evidence of recurrence.

This case shows that endoscopic cauterization of the fourth branchial cleft sinus can be considered as a less invasive effective alternative procedure to open excision.

Most congenital lateral cervical cysts, fistulae, and sinuses are considered to originate from the branchial apparatus. Branchial cysts have no external or visceral opening and thus retain secretions, whereas branchial sinuses communicate with either the skin or the pharyngeal lumen, and fistulas are tracts connecting the pharynx to the skin.²

The reported prevalence of fourth arch anomalies is very low. According to most authors, the number of published cases is less than 100, representing 1–4% of all branchial anomalies.²

Anomalies in the embryogenesis of the fourth branchial apparatus were first reported in 1972.¹

Fourth arch anomalies were usually located on the left (94%) and generally presented as acute suppurative thyroiditis (45%) or recurrent neck abscess (42%).²

The left side predominance might be because of the more complex and extended pathway of the fourth branchial tract on the left side. It might also be because of the absence or involution of the ultimobranchial body on the right side.²
We present a case of the fourth branchial cleft sinus that was treated by endoscopic cauterization with a 23 month period of follow up.

2. Case presentation

A 15 year old female presented to our emergency department with a 5 day history of a mass on the left side of the neck associated with dysphagia, restriction of neck movement and subjective fever with no history of discharge or respiratory distress.

The patient had 6 previous similar attacks in the past 2 years for which she had been managed in every incident by antibiotics and incision and drainage.

In the past medical history the patient was followed up by the endocrinology department as a case of recurrent suppurrative thyroiditis with normal thyroid function test.

On examination; we noted a 4 × 5 cm hot tender immobile mass on the anterior left side of the neck. ENT examination otherwise was unremarkable.

The CT scan with contrast revealed a hypodense area on the left side of the neck of about 3 × 4 cm with ring enhancement (Fig. 1).

The abscess extends from the level of the glottic area down to the thyroid level displacing the left thyroid lobe with no airway compromise with evidence of reactionary changes involving the subcutaneous tissue and sternocleidomastoid muscle (Fig. 2).

The patient was admitted as a case of neck abscess and underwent incision and drainage under general anesthesia and microlaryngoscopy plus examination of pyriform fossae which was decided to rule out the differential diagnosis of fourth branchial cleft anomaly based on the above mentioned history of recurrent neck infection with recurrent thyroiditis and the radiological imaging.

Endoscopic examination revealed an opening in the apex of the ipsilateral left pyriform fossa (Fig. 3).

The patient was kept in the hospital on an I.V. antibiotic (clindamycin) with the impression of infected fourth branchial cleft sinus.

Three days later the patient was scheduled for microlaryngoscopy.

On the operating table under endotracheal anesthesia, microsuspension was performed exposing the left pyriform sinus and locating the sinus opening (Fig. 3).

Fogarty balloon catheter was passed through the identifiable sinus opening, the balloon then inflating and the sinus dilated (Fig. 4A).

Cautery was then passed through the sinus opening (Fig. 4B) then cauterization was done on low power until the surrounding tissue got shrunk and blanched (Fig. 4C).

Cautery was then withdrawn partially and subsequent cauterization was done at the level of the opening (Fig. 4D).

Postoperative stay was uneventful.

The patient was followed up regularly in the outpatient department for 23 months with no evidence of recurrence.

3. Discussion

Anatomically, fourth branchial arch anomalies represent vestiges of a tract that originates as a sinus tract from the apex...
of the pyriform sinus. It travels inferiorly in the tracheoesophageal groove, posterior to the thyroid gland, and into the thorax, where it loops below the aorta on the left and subclavian artery on the right. The tract then changes direction, coursing superiorly, passing posterior to the common carotid artery, and ascending in the neck to reach the hypoglossal nerve. It then makes a second loop around the hypoglossal nerve and finishes its course at the medial border of the sternocleidomastoid muscle. The descending part of this tract before the first loop is the most common location of clinical infection.2

It is difficult to differentiate fourth from third branchial arch anomalies because both sinuses are expected to open into the pyriform sinus. The differentiation between the two conditions lies in determining the relationship of the sinus to the superior laryngeal nerve, which can only be identified with surgical exploration. If the sinus passes below the superior laryngeal nerve, a fourth branchial pouch sinus is suggested, whereas if the sinus passes above the superior laryngeal nerve, a third branchial pouch sinus is suggested. Endoscopic clues to the origin of a sinus can be gained from the location in the pyriform sinus. Openings in the apex of the piriform sinus suggest fourth branchial cleft anomalies. The findings of pathologic examination of the excised specimen to determine the pouch of origin can be confusing. The presence of the parathyroid tissue and thymus in a specimen suggests that the sinus is of third pouch origin, as both the inferior parathyroid glands and the thymus are of third pouch origin. The presence of the parathyroid and thymus tissue can be misleading, however, because parathyroid tissue may be a part of the superior parathyroid gland (a fourth pouch structure) and an accessory thymus can develop from the fourth pouch, making these findings less specific for a third branchial cleft sinus.3

The presentation can be classified into two forms, the neonatal form manifests as a very large cervical mass that may or may not be infected and readily compresses surrounding structures. The mass may cause dyspnea with stridor and feeding difficulties. The second form of the disease presents in later childhood as a cervical abscess associated with local clinical features.4

Cases of mediastinal abscess of the fourth branchial arch origin have been reported and must be considered. Acute suppurative thyroiditis is a late clinical presentation, suggesting that the sinus tract may progress with age, beginning close to the pyriform sinus at a younger age and migrating lower in the neck in older children.2

A combination of ultrasound, computed tomography (CT) with or without oral contrast, barium swallow study looking for the sinus tract, thyroid scan, or magnetic resonance imaging (MRI) may aid in the diagnosis.4 However, the best diagnostic examination is endoscopy, which allows confirmation of the diagnosis by direct visualization of the orifice of the tract in the pyriform fossa and the Betz fold.5

The classic management described in the literature is surgical resection using a cervical approach. However, recently a less invasive treatment has evolved, namely, endoscopic cauterization limited to the sinus tract orifice.1,2,4,5

Curative procedure should only be attempted in the absence of acute infection.1,2,4

A number of endoscopic techniques have been reported in the literature. Verret et al. have described the introduction of
a balloon catheter for the dilation of the sinus tract orifice to allow endoscopic cauterization. Electrocoagulation was also used by Jordan et al. with the aid of a diathermy probe. Alternatively, Sayadi et al. used a low-power diode laser, while 2 other groups used chemical cauterization with trichloroacetic acid. Kim et al. and Pereira and Smith performed chemical cauterization by introducing a stick of silver nitrate for 3-4 s into the sinus tract and at the sinus tract orifice.\(^5\)

Endoscopic treatment represents a minimally invasive technique using cauterization to obliterate the internal opening of a pyriform sinus tract during a quiescent period. It can be done as a definitive management or along with the surgical excision of the fourth branchial anomaly.\(^2\)\(^,\)\(^5\) Possible advantages of this technique over open neck surgery include a lower complication rate with similar rates of recurrence as open neck surgery.\(^2\) Recurrences can be treated by repeated cauterization using the same technique, with good long-term outcomes.\(^5\)

In our case report we present a patient with a fourth branchial cleft sinus anomaly presented as recurrent thyroiditis with recurrent neck abscess who was successfully managed by a single endoscopic cauterization of sinus tract after initial incision and drainage of neck abscess with antibiotics coverage with no evidence of recurrence in a 23 month period of follow up.

We recommend endoscopic examination as a routine investigational tool in all patients who present with neck mass, recurrent neck infection and recurrent thyroiditis looking for evidence of sinus opening in pyriform fossa.

Definitive management should be done on the absence of acute infection.

The main aim of cauterization was the closure of the sinus orifice.

**References**