

## Rare Case of Coexisting First and Second Bilateral Branchial Fistulas

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### Abstract

Branchial arch anomalies are one of the most common congenital anomalies that are usually unilateral, bilateral presentation is rare. We describe what we believe a rare reported clinical case of a 34-year-old man with coexisting first and second bilateral branchial fistulas associated with no family history or associated syndrome.

**Keywords:** Bilateral; Branchial; First Arch; Second Arch; Fistula

### Introduction

A branchial anomaly is a congenital developmental defect that arises from the primitive branchial apparatus [1]. The majority of children's cervical neck masses are congenital in nature. Twenty percent of these congenital neck masses are branchial anomalies [2]. The development of many structures in the head and neck is intimately related to branchial arches, which are transient embryonic structures of muscle, bone, and other similar mesodermal derivatives seen in the head and neck. Bilateral presentation of branchial anomalies is rare, and is seen in about 2-3% of all cases [3,4]. Mostly, such cases have a familial component.

We will describe the case of a 34-year-old man, who presents a bilateral branchial fistula due to the association of first and second branchial cleft.

### Case Report

A 34-year-old man presented to the ENT department with bilateral pinpoint in the lower one-third of the neck with intermittent discharge from right side since birth. The patient reported having these small openings since birth, with episodes of infection, inflammation and occasional fluid leaks. The amount of secretions would increase during episodes of upper respiratory tract infections and decrease with antibiotic therapy. Further history was non-contributory.

On examination (Figures 1,2 and 3) 0.2×0.2 cm size openings were present at the junction of the upper two-third and lower one-third of the anterior border of sternocleidomastoid muscle (SCM), bilaterally. Examination also revealed 0.3×0.2 cm asymptomatic openings in front of both ear. The auricular pavilion was of normal shape, size and position, and the eardrum was normal. Further, clinical examination was normal.



**Figure 1:** Left side view showing pin-point opening in the preauricular area and cyst in the neck on left side



**Figure 2:** Frontal view showing pin-point opening in the neck on left side



**Figure 3:** Surgical excision

Cervical ultrasound had shown: 3.5 cm right fistular tract extending from the junction of the upper two-third and lower one-third of the anterior border of sternocleidomastoid muscle to the right palatine tonsil.

Associated to 2.5 cm left fistular tract extending from the junction of the upper two-third and lower one-third of the anterior border of sternocleidomastoid muscle towards the left cervical axis

Abdominal ultrasound was and renal examination revealed normal renal function, excluding the possibility of a branchio-oto-renal syndrome.

Audiometry was normal. The patient was operated under general anesthesia; his fistulas were excised until their origin.

An incision made of orange in the face of the fistula, dissection following the fistulous pathway. Taking off the cyst from the deep structures, that is to say the X, XI, XII and its descending branch, vein internal jugular and carotid vessels, cyst removed in totality, closure plane by plane.

The patient's postoperative course was uneventful and remained symptomless at one-year follow-up.

## Discussion

The gill arches develop between the fourth and seventh week of gestation and form the embryological precursors of the ear and muscles, blood vessels, bones, cartilage and mucous membranes of the face, neck and pharynx. In total, six pairs of branchial arches are formed on either side of the pre-pharyngeal intestine in the cranio-caudal succession. The fifth pharyngeal arch is usually only rudimentary or never formed, so five arches eventually form adult structures [5,6]. Schematically, the sixth arch is often depicted as part of the fourth arch because of its small size.

The development and differentiation of the primitive branchial apparatus occurs during the second and seventh weeks of gestation [7]. Initially, five ridges appear on the ventrolateral surface of the embryonic head. These ridges form the gill arches. Each arch consists of a mesenchyme nucleus and is externally covered with ectoderm, representing the branchial cleft, and internally with endoderm, representing the gill pocket. These slots and pockets come together to form a closure membrane. Invasion of the surrounding mesenchyme gradually clears the cleft and pocket in humans [8].

Anomalies of the gill arches may occur in the first, second, third or fourth arches. First arch constitutes 8-10% of branchial anomalies [9]. We described two types of first branchial anomalies. In type I anomalies, there is a duplication of the external auditory canal, which open in the preauricular area. In type II anomalies, the tract is in close association with the parotid gland and the facial nerve. They open posterior or inferior to the angle of mandible [10]. It is sometimes difficult to decide whether these

pre-auricular sinuses and cysts are inclusion dermoids resulting from epithelium trapped between the developing auricle tubercles or remnants of the first branchial groove epithelium that have failed to reabsorb.. However, if the pre-auricular sinuses do arise from a lack of fusion of the six hillocks of His, then such sinuses are usually very small and present as pits or blind superficial sinus[11,12].

The second pharyngeal pouch develops in turn in the tonsils. Thus, anatomically, the second branchial fistula typically develops at the anterior edge of the sternocleidomastoid muscle, the fistular orifice being located at the junction of the middle and lower thirds of the muscle. The tract usually pierces the platysma and climbs along the carotid sheath through the carotid bifurcation (superficial to internal carotid artery) and passes through the hypoglossus and glossopharyngeal nerves. The characteristic leaflet extends upwards, ending in the upper half of the posterior tonsillar pillar, in the supratornsary fossa, or directly on the surface of the tonsils [11,13].

An appreciation of the related embryology is crucial to understanding the surgical management and associated risks [10].

We describe the case of young man in our ENT department with a bilateral fistula of the second branchial arch associated with a preauricular bilateral fistula confirmed with ultrasound. The diagnosis of this abnormality is generally based on the clinical and Para clinical characteristics described which allowed eliminating other differential diagnoses.

The treatment protocol for such lesions is surgical excision; antibiotics are only used to treat infections in the tract. Knowledge of embryology is crucial for understanding the surgical management and associated risks.

A rigorous clinical and Para clinical analysis of cystic malformations and their fistulous evolution is necessary.

Surgical excision should include dissection of the fistulous tract along its entire length of the tract with complete excision of the cyst.

## Conclusion

The presentation of first and second arch bilateral anomalies is very rare. To the best of our knowledge, only few cases with similar findings have been reported. These cases must be managed and a thorough investigation must be conducted to rule out other congenital anomalies and other associated malformations.

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