

# Unusual coexistence of first and second branchial fistulas: clinical case and review of the literature

Journal of International Medical Research

48(8) 1–6

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DOI: 10.1177/0300060520944303

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## Abstract

Branchial fistulas are uncommon in the clinical setting. The coexistence of first and second branchial fistulas has not been previously reported. We herein describe a 12-year-old girl who presented with a 2-year history of repeated swelling and purulence behind the right earlobe and neck. According to the patient's physical and auxiliary examination findings, she was diagnosed with coexisting first and second branchial fistulas, both of which were completely removed by surgery. No clinical signs of fistula recurrence were present at the patient's 20-month postoperative follow-up. Ipsilateral coexisting first and second branchial fistulas are very rare; thus, a false-positive diagnosis can easily occur if the doctor does not carefully perform specialized physical examinations. Surgery is an effective method for treating this condition. Adequate preoperative imaging preparation is imperative to ensure the most effective course of treatment. The purpose of this article is to improve clinicians' awareness of this disease, thereby effectively reducing the rates of missed diagnosis and recurrence.

## Keywords

Branchial fistula, preoperative imaging, surgery, complete resection, secondary infection, case report

Date received: 6 April 2020; accepted: 1 July 2020

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## Introduction

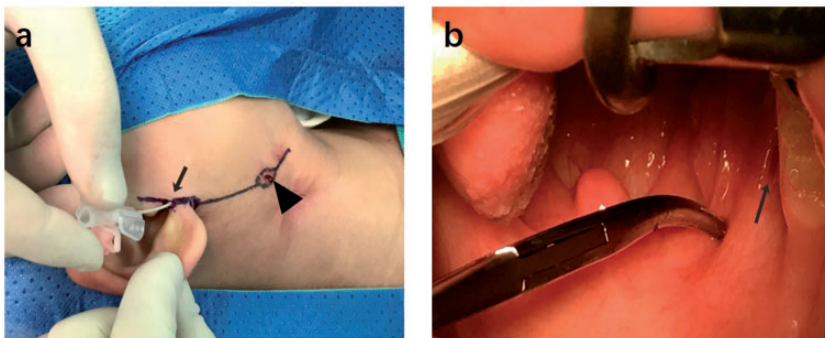
A branchial fistula is a congenital fistula associated with abnormal development of the branchial apparatus, which is located in the laterocervical region. It is infrequently encountered in the clinical setting. Most branchial fistulas originate from the second branchial groove; they rarely develop from the first, third, or fourth branchial groove. Bilateral branchial fistulas are occasionally seen<sup>1</sup>; however, the coexistence of two types of branchial fistulas on the same side is extremely rare. We herein report a case of ipsilateral coexisting first and second branchial fistulas in a patient who presented to our department.

## Case presentation

A 12-year-old girl presented with a 2-year history of repeated swelling and purulence behind the right earlobe and neck. The patient reported no earache, otorrhea, hearing loss, halitosis, fever, cough, or swallowing discomfort. She visited a local hospital and was diagnosed with a second branchial fistula associated with infection. The redness and swelling of the neck could not be completely cured by antibiotics.

## Physical examination

Upon presentation to our hospital, the skin behind the right earlobe was congestive and scabbed. A small hole was present at the right incisura intertragica (Figure 1(a)). The fistula extended to the deep surface of the parotid gland. Another fistula was seen behind the lower aspect of the right mandibular angle and at the leading edge of the middle segment of the sternocleidomastoid muscle (Figure 1(a)). The fistula coursed upward and forward (i.e., inward). Another internal fistula was found above the right pharyngopalatine arch (Figure 1 (b)). Contrast agent was injected into the external fistula and an enhanced computed tomography scan was performed. The fistulas were examined on the computed tomography images (Figure 2(a) and (b)). Contraindications were excluded before surgical treatment, which was performed under general anesthesia. During the operation, the upper segment of the fistula was found to be in close contact with the cartilage of the external auditory canal. The fistula was explored successively in the parotid parenchyma, the deep surface of the main facial nerve, and the superficial surface of the posterior belly of the digastric muscle. The fistula then diverged at the lower neck



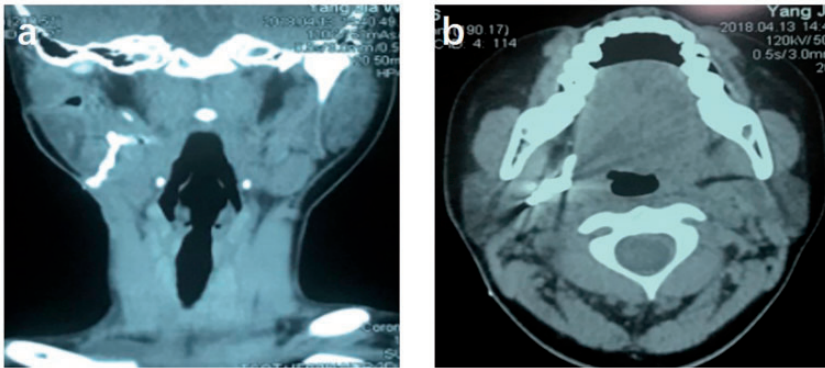
**Figure 1.** Intraoperative photographs. (a) Internal fistula at the ear (arrow) and external fistula at the neck (arrowhead). (b) Internal fistula at the oropharynx (arrow).

behind the mandibular angle. The lower end extended into the external neck fistula, and the deep part advanced forward and inward through the surface of the vagina vasorum, the stylohyoid muscle, and the superficial surface of the hypoglossal nerve (Figure 3). This fistula was found to end in the oropharynx. The fistula was completely removed (Figure 4), and no complications such as facial paralysis were observed.

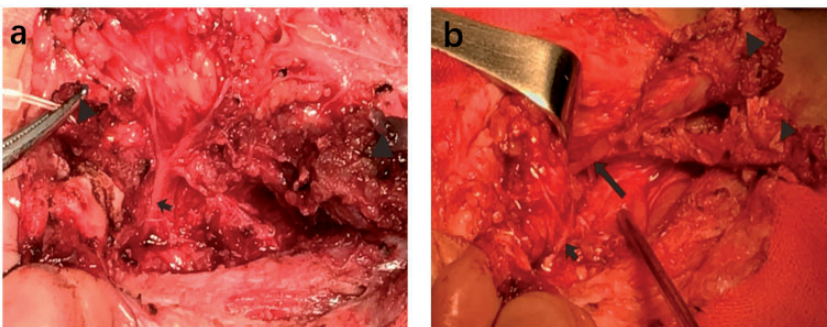
## Discussion

Most branchial fistulas are considered to be associated with incomplete closure of the branchial groove and rupture of the closed

membrane. The first branchial apparatus mainly develops into the maxilla, mandible, Eustachian tube, cavum conchae, external auditory canal, and tympanic membrane epithelium. The second branchial apparatus mainly forms the facial muscles, styloid process, and auricle. Dysplasia of the branchial apparatus is mainly related to these structures. First branchial fistulas are anatomically divided into type I and type II.<sup>2</sup> Fistulas have internal and external orifices, and the internal orifice of most first branchial fistulas are located in the external auditory canal (bone or cartilage) and in the tragus, although some are located in the Eustachian tube.<sup>3</sup> The fistula is usually



**Figure 2.** Computed tomography images of fistula. (a) Coronal view. (b) Horizontal view.



**Figure 3.** Surgical exploration. (a) The triangular arrowheads indicate the internal fistula at the ear (left) and external fistula at the neck (right). (b) The short arrow indicates the facial nerve trunk. The long arrow indicates the fistula that extended into the oropharynx.



**Figure 4.** Gross specimen. Pathologic examination showed that the epithelium in the fistula was continuous.

**Table I.** Common internal and external orifices associated with first and second branchial fistulas.

Fistula types	Internal orifices	External orifices
First	External auditory canal Eustachian tube	Anterior margin of the SCMM before or after the mandibular angle
Second	Tonsillar fossa	Between the middle and lower third of the anterior margin of the SCMM

SCMM: sternocleidomastoid muscle.

located behind the parotid gland or courses through the parenchyma of the parotid gland and then passes through the deep or shallow surface of the facial nerve to the external orifice. External fistulas are usually located in the anterior margin of the sternocleidomastoid muscle before or after the mandibular angle. The external orifice of most second branchial fistulas is located at the junction between the middle and lower third of the anterior margin of the sternocleidomastoid muscle. It usually passes between the internal and external carotid arteries and reaches the internal fistulas of the tonsil fossa via the superficial surface of the hypoglossal nerve, glossopharyngeal nerve, and stylohyoid ligament (Table 1). In the present case, pathological examination showed that the inner wall of the branchial fistula was lined with squamous

epithelium. In addition to symptoms and physical examination findings, the diagnosis of branchial fistulas should also be assisted by imaging examination. Before the examination, a contrast agent (e.g., iodide oil, gadopentetate dimeglumine) should be injected through the fistula, and X-ray or computed tomography examination should then be performed to determine the direction of the fistula; this can greatly improve the success rate of the operation. Treatment is both surgical and nonsurgical. Surgery is the main treatment method, and radical removal of the fistula is the key to a successful outcome. Preoperative local injection of methylene blue<sup>4</sup> and endoscopic-assisted positioning of the internal fistula can improve the success rate of the operation to a certain extent. Nonsurgical methods usually include the

use of corrosive drugs such as trichloroacetate,<sup>5</sup> which have an unstable effect. Such drugs are not commonly used.

Congenital fistulas of the neck are rare. The annual incidence of first branchial fistulas is reportedly 1 per 1,000,000 individuals, and the incidence is higher in women than men.<sup>6</sup> A branchial fistula with a thyroglossal duct cyst has been occasionally reported in the literature.<sup>7</sup> The coexistence of two types of branchial fistulas on the same side has been rarely reported. In this case, the patient had a history of recurrent neck swelling. Although she was admitted to many hospitals, no surgery was performed, likely for the following three reasons. First, the neck infection was not well controlled. Second, branchial fistulas are complicated and have a high incidence of surgical complications. Third, the patient and her family had high expectations and were worried about possible complications of surgery. Regarding the first point, antibiotics should be administered according to the results of bacterial culture and drug sensitivity. The drug dose and treatment course should be sufficient. Drainage of the abscess cavity and fistula should be conducted simultaneously. In addition, cleaning of the internal fistula and local microwave physiotherapy should be carefully performed. In our patient, the inflammation was finally controlled to a satisfactory degree, mainly because of sufficient local antibiotic irrigation of the fistula. Regarding the second point, after the inflammation has been fully controlled, angiography and imaging examination should be performed to determine the route and extent of the fistula. The clinician should carefully read the film before the operation and determine the surgical incision based on the results of the physical and auxiliary examinations. Meanwhile, possible problems that might be encountered during the operation should be fully estimated, and countermeasures should be

taken. The operator's knowledge of the relevant anatomy should be proficient, and the intraoperative movements should be gentle to avoid damage to the relevant structures. Thorough knowledge of the anatomy of the facial nerve is a key aspect of first branchial fistula resection.<sup>8</sup> Methylene blue can be used for preoperative fistula angiography. During the operation, the fistula tissue can be traced according to the angiographic findings, and some of the skin and cartilage can be removed if necessary to avoid recurrence.

## Conclusions

Because of the complicated surgical treatment of branchial fistulas, especially complex fistulas, perioperative management should be thorough and the surgical method should be adjusted according to the specific type of fistula. Complete resection of the fistula is the key to preventing recurrence because the second operation will be exceptionally difficult.

## Acknowledgement

The authors thank the patient's parents for providing informed consent to publish her case and related images.

## Authors' contributions

DQL and GWP were responsible for the study conception and design. DQL and GQL drafted the initial manuscript and revised it critically for important intellectual content. JQ and JYW collected and organized the photographs. All authors read and approved the final manuscript.

## Availability of data and materials

The data sets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

## Declaration of conflicting interest

The authors declare that there is no conflict of interest.



### Ethics and informed consent

No animal or human experiments were performed in this study; therefore, ethics committee approval was not required. Given that the patient was 12 years old, written informed consent for publication of the patient's clinical details and clinical images was obtained from the patient's parents.

### Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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