

First Branchial Cleft Fistula Piercing through the Main Trunk of the Facial Nerve

Masazumi Moriyama, MD*†
 Kosuke Kuwahara, MD, PhD*†
 Masahiro Nakagawa, MD, PhD†
 Hideaki Kamochi, MD, DDS,
 PhD*

Summary: First branchial cleft fistulas are congenital malformations that result from the incomplete closure of the ectodermal portion of the first branchial cleft. These fistulas typically appear as small pits or subcutaneous masses in the upper neck and cheek and can cause pain due to infection and inflammation. Surgical excision is the most effective treatment, but special attention is necessary to avoid facial nerve injury due to the proximity of the lesion to the nerve and variations in their arrangement. Here, we report the successful treatment of a first branchial cleft fistula piercing through the main trunk of the facial nerve in a 3-year-old girl. Intraoperative findings revealed that the fistula in the parotid gland opened into the cheek area from the ear canal. Identification of the facial nerve trunk was challenging due to the malformation of the lower end of the auricular cartilage, which is an anatomical landmark of the facial nerve. The trunk of the facial nerve was divided proximally by the fistula and merged just past the fistula. Preoperative magnetic resonance is important for determining the fistula location, surrounding anatomical variations, and fistula-facial nerve arrangement. Furthermore, early surgical treatment should be considered to prevent tissue scarring and adhesion due to infection, which can lead to facial nerve injury. (*Plast Reconstr Surg Glob Open* 2023; 11:e5173; doi: 10.1097/GOX.0000000000005173; Published online 7 August 2023.)

First branchial cleft fistulas are congenital malformations that originate from incomplete closure of the ectodermal portion of the first branchial cleft. They are rare, accounting for 8%–10% of all branchial cleft anomalies. They typically appear as small pits or subcutaneous masses in the upper neck and cheek and can cause pain due to infection and inflammation. Surgical excision is the most effective treatment, but special attention must be taken to avoid facial nerve injury due to the proximity of the lesion to the nerve and variations in their arrangement.¹ In this report, a case of a first branchial cleft fistula piercing through the main trunk of the facial nerve is presented.

CASE PRESENTATION

A 3-year-old girl presented to our department with a history of recurrent pain and discharge from a pit located

in the left cheek for 2 years (Fig. 1). The patient had received local treatment with general antibiotic therapy without improvement. Otoscopy showed a small dimple and discharge on the floor of the left ear canal.

Contrast-enhanced magnetic resonance imaging (MRI) showed a fistula in the parotid gland opening into the cheek area from the ear canal (Fig. 2). The fistula was thick, and its walls were much thicker than those of an auricular fistula. Furthermore, the fistula was attached to the facial nerve and split proximally from the trunk of the facial nerve (Figs. 3 and 4). Because there was no active infection, surgical excision was performed as soon as possible to prevent tissue scarring and adhesion due to recurrent infection.

Surgical excision was performed under general anesthesia using the parotidectomy approach. Intraoperative findings showed that the upper end of the fistula was integrated with the ear canal cartilage, and the lower end of the auricular cartilage (pointer cartilage: anatomical landmark of the facial nerve) was malformed. Due to the lack of a landmark, identification of the facial nerve trunk was difficult and required an approach from the periphery of the nerve. The trunk of the facial nerve was divided proximally by the fistula and merged just past the fistula, as if the fistula was piercing through the main trunk of the facial nerve. Because the wall of the fistula was hard and had no adhesion to surrounding tissue, nerve-preserving excision was easy.

From the *Department of Plastic Surgery, Shizuoka Children's Hospital, Shizuoka, Japan; and †Department of Plastic and Reconstructive Surgery, Hamamatsu University School of Medicine, Hamamatsu, Japan.

Received for publication June 4, 2023; accepted June 23, 2023.

Copyright © 2023 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

DOI: 10.1097/GOX.0000000000005173

Disclosure statements are at the end of this article, following the correspondence information.



Fig. 1. Patient with recurrent discharge from pit located in left cheek.

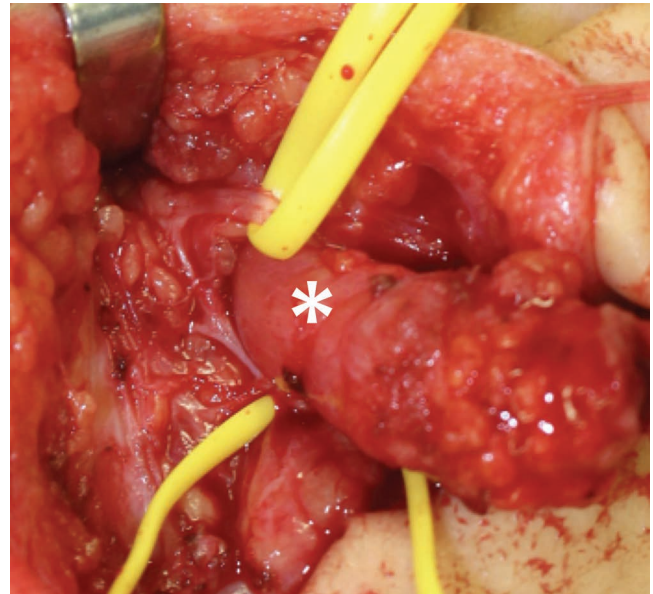


Fig. 3. Surgical site. The asterisk (*) indicates the first branchial cleft fistula contacted with the branch of facial nerve.

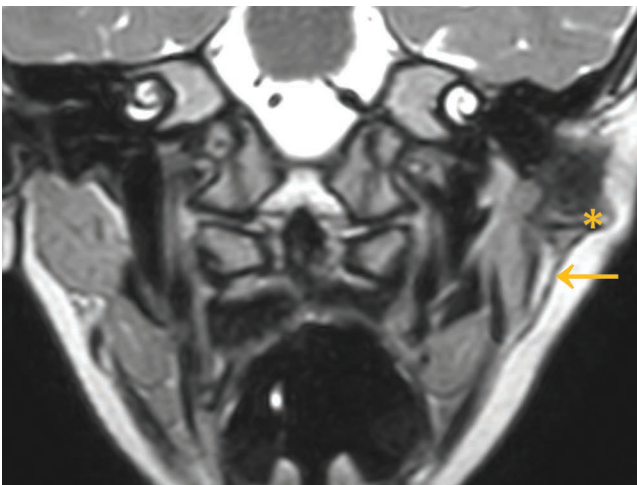


Fig. 2. MRI demonstrating the radiological characteristics of the first branchial cleft fistula (←) in the left parotid space extending external auditory cartilage (*).

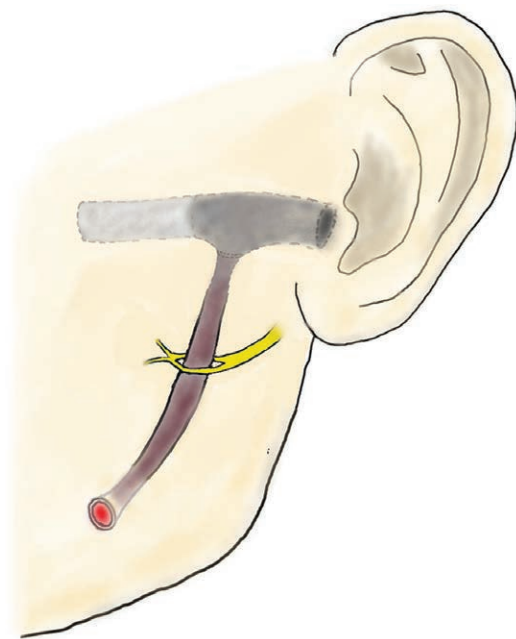


Fig. 4. The schema of the branchial cleft fistula piercing the main trunk of the facial nerve.

On histological examination, the fistula was a first branchial cleft anomaly. During the 12-month follow-up period, there were no signs of clinical recurrence or facial nerve palsy. The surgical scar has matured, and there is no constriction of the ear canal.

DISCUSSION

In general, first branchial cleft anomalies are caused by incomplete closure of the ectodermal portion of the cleft and account for only 8% of all branchial cleft anomalies.

According to Olsen,² 68% of these anomalies are cysts, 16% are sinuses, and 16% are fistulae. Sinuses and fistulae are more commonly encountered in infants and children, whereas cysts are more common in adults.³

Various classifications have been proposed to describe first branchial cleft anomalies. Belenky's classification subdivides the relationship between the fistula and the facial nerve into three types based on anatomical features, which can be helpful for surgical treatment.⁴

First branchial fistulae, such as the one observed in this case, that pierce through the main trunk of the facial nerve are quite rare and do not fit into any of the existing classifications. Crymble and Braithwaite reported a similar case in 1964.⁵ In these cases, it was most difficult to remove the tract without damaging the nerve. Some points related to treatment are presented.

First, evaluating the proximity of the fistula to the facial nerve is crucial for treatment. MRI is invaluable for predicting the relationship between first branchial cleft anomalies and the facial nerve, although it cannot define the peripheral branches of the facial nerve.⁶ Therefore, intraoperative findings are the most important for safely identifying and preserving the facial nerve.

Second, early surgical treatment is desirable to avoid repeated infections and incisional drainage, which can cause adhesions and scar formation between the fistula tract and surrounding tissues, such as nerves.⁷ Therefore, surgery should be performed as soon as possible for safe treatment.

Finally, it is important to identify any other anatomical malformations besides the fistula. Zhang described malformations of the external auditory canal and middle ear.⁸ In the present case, the lower end of the auricular cartilage (the pointer cartilage, an anatomical landmark of the facial nerve) was malformed. Due to the lack of a landmark, identifying the facial nerve trunk was difficult and required an approach from the periphery of the nerve.

CONCLUSIONS

In the present case, the first branchial cleft fistula pierced through the main trunk of the facial nerve. Careful preoperative evaluation and early surgical treatment are important for safe excision while preserving the

facial nerve. Identification of any additional anatomical malformations beyond the fistula is also crucial for ensuring successful surgery.

Masazumi Moriyama, MD

Department of Plastic Surgery

Shizuoka Children's Hospital

860 Urushiyama, Shizuoka city

Shizuoka 420-0953, Japan

E-mail: prs.moriyama@icloud.com

DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

REFERENCES

1. Chen Z, Wang Z, Dai C. An effective surgical technique for the excision of first branchial cleft fistula: make-inside-exposed method by tract incision. *Eur Arch Otorhinolaryngol.* 2010;267:267–271.
2. Olsen KD, Maragos NE, Weiland LH. First branchial cleft anomalies. *Laryngoscope.* 1980;90:423–436.
3. Chaouki A, Lyoubi M, Lahjaouj M, et al. Atypical first branchial cleft fistula. *Int J Surg Case Rep.* 2021;78:159–161.
4. Belenky WM, Medina JE. First branchial cleft anomalies. *Laryngoscope.* 1980;90:28–39.
5. Crymble B, Braithwaite F. Anomalies of the first branchial cleft. *Brit J Surg.* 1964;51:420–423.
6. Lendoye W. An extremely rare case of adult with first branchial cleft fistula. *Ann Med Surg.* 2021;70:102807.
7. Jang M, Dai C. An effective surgical approach for the management of recurrent first branchial cleft fistula with cysts: a case report. *J Int Adv Otol.* 2017;13:419–421.
8. Zhang CL, Li CL, Chen HQ, et al. First branchial cleft cyst accompanied by external auditory canal atresia and middle ear malformation. *World J Clin Cases.* 2020;8:3616–3620.