Diagnosis and management of first branchial fistula: a study of 12 cases

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Abstract

Objectives To report authors’ experiences in the diagnosis and treatments of congenital first branchial fistula (congenital auriculocervical fistula). Materials and Methods Twelve cases of congenital first branchial fistula were reviewed. Of these, 8 underwent fistulectomy with facial nerve dissection and partial parotidectomy and 4 underwent simple fistulectomy. Results The inner openings (upper opening) of fistulae lay in the following sites: inferioposterior wall at the junction of cartilaginous and bony segments of the auricular canal and inferior wall of cartilaginous auricular canal. The outer openings (lower opening) lay along the anterior border of upper sternocleidomastoid muscle, at the mastoid tip and posterior to the mandibular angle. Complete fistulae resection was achieved in all but one case. Eleven cases were followed for 5 year with no recurrence. Recurrence occurred in 1 case 6 months after the primary surgery and revision surgery was performed. Conclusions Pre–operative radiography for the location and course of the fistula is crucial for successful fistula resection, especially in cases with past infections. Facial nerve dissection should be done routinely for deeply located fistulae.

Key words Congenital Malformation; First Branchia; Auriculocervical Fistula; Diagnosis; Surgery

Introduction

The congenital first branchial fistula, one of branchial deformities, is also named as congenital auriculocervical fistula from the location of its openings. The first branchial deformity is thought to develop as a result of incomplete obliteration of the branchial cleft between the mandibular process of the first and second arch. It may present as a cyst, sinus or fistula.

The congenital first branchial fistula is rare and often misdiagnosed. The difficulty in treating this disease relates to its close relations to the facial nerve and parotid gland. In the past years, we treated 12 patients with the first branchial fistula.

These cases are reviewed in this report in the context of the diagnosis and surgical treatment of this condition.

Patients and Methods

Clinical data

Twelve patients were diagnosed with first branchial fistula and treated at the Department of Otorhinolaryngology and Head–Neck Surgery, the Second Affiliated Hospital, Sun Yat-sen University, Guangzhou, China, between 1980 and 2003. Seven patients were male and 5 were female. The age ranged from 8 to 38 years (mean = 15 years). Presenting symptoms included fistulae with/without suppurative drainage in the postauricular area or submandibular triangle (n = 4) and painful masses in the postauricular area or neck (n = 8). The inner opening was located in the external auricular canal in all 12 cases. In 6 cases, the diagnosis was
established by the authors at the first visit. The rest 6 cases were misdiagnosed as postauricular fistula, postauricular abscess, otitis media, lymphatitis, sebaceous abscess or atheroma of auricular lobule at other facilities before coming to the authors. All cases had iodized oil roentgenograph or CT scan of the fistula (Figure 1). Imaging was not successful in 1 case due to heavy pressure and imaging findings were inconsistent with surgical findings in another case.

Surgical management

All patients were treated surgically, 2 under local anesthesia and the rest 10 under general anesthesia. Methylene blue injection into the fistula was applied in 9 cases immediately before operations, but was helpful only in 4 cases in guiding fistulae dissection due to scar formation related to previous infections in the rest cases.

Simple fistulectomy was possible in 4 cases, with longitudinal incision, clean dissection and complete resection of the fistula together with the inner opening. A drainage film was left in the wound for 24 hours. Facial nerve dissection (FND) and/or partial parotidectomy were necessary in 8 cases.

In these case, a “Y” incision around the auricular lobule or a parotidectomy incision was used. The auricular lobule was lifted and the parotid exposed. The main trunk and branches of the facial nerve were dissected (Figure 2). The fistula was located lateral to the facial nerve in 5 cases and medial to the main trunk in 3 cases. In 1 case, the fistula showed branched structures and extended deep to the skull base (petrous apex).

The fistula was carefully separated from the facial nerve, with partial parotidectomy when necessary, and traced to the external auricular canal for complete resection with its inner opening. The external auricular canal was packed with iodoform gauze dressing. A “Y” drainage tube with suction was used for 48 hours post-operatively.

Results

Complete fistula resection was achieved in 11 cases with no recurrence during the 5 year plus follow–up period. One fistula with complex branches and extension to the skull base was not well demonstrated on pre–operative iodized oil roentgenograph and relapsed 6 months later. A revision surgery was performed in this case (table1). Post–operative draining parotid fistula occurred in 1 case with facial nerve dissection and partial parotidec-
tomy, which was successfully treated with pressure bandage and two weeks of atropine. Post-operative facial weakness was seen in 1 case which recovered in 4 months. Histological examination of specimens confirmed the diagnosis of fistula in all cases.

Discussion

Diagnosis of the congenital branchial fistula

Congenital branchial fistulae are rare in the clinic, consisting of only 1–8% of branchial deformities. It usually has two openings: outer and inner. The outer opening may lie in one of the following sites: lateral side of the upper neck, behind the mandibular angle and at the mastoid tip. The inner opening may lie in the external auricular canal, tympanic cavity or Eustachian tube. This disease is often misdiagnosed in the clinic, because it is uncommon and physicians are not familiar with it. Operations on fistula, cyst or abscess based on incorrect diagnoses further increase difficulties for future treatment. Six of our 12 cases were misdiagnosed as postauricular fistula/abscess, otitis media, lymphatitis, sebaceous abscess or atheroma of auricular lobule in other clinics. One of the misdiagnosed cases had been operated upon more than ten times before seen by the authors. First branchial fistula should be considered when a fistula or abscess is in the area higher than the hyoid, anterior to the sternocleidomastoideus, posterior to the

<table>
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<th>side</th>
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mandibular angle or in the external auricular canal. The external auricular canal, especially the posterior–inferior area at the junction of the cartilaginous and osseous segments, should be carefully examined. Presence of the inner opening of the fistula strongly indicates the diagnosis of first branchial fistula.

In addition to physical examination, imaging studies are also an important part of the diagnostic work up. Iodized oil roentgenograph involves injecting 40% iodized oil into the outer opening for imaging of the location and course of the fistula. This is helpful for both diagnosis and planning of surgery. All 12 cases in our series received iodized oil roentgenograph and images in 11 cases were consistent with surgical findings.

The fistula can be classified based either upon its anatomy or pathology. The authors believe that classification should reflect the fistula’s relations to the facial nerve and/or parotid gland, as this is critical in planning surgical approaches. During development, the facial nerve is derived from the second branchial arch. The second branchial arch later covers the first branchial arch as a result of its rapid development. So the relations between the facial nerve and fistula need to be carefully studied during the diagnostic process.

The relations between the fistula and skull base are also important. In 1 case in our study, a branch of the fistula extended deep to the skull base and was not well demonstrated on pre-operative imaging. It surprised the surgeon during the surgery and may have led to incomplete resection and later recurrence.

Surgical management of congenital branchial fistula

Surgery is the only way to treat fistula. Operation should be considered after acute infection has been well controlled. In our reported cases, 3 presented with acute infection and abscess. These were treated with abscess drainage and antibiotics before fistula removal was attempted. Methylene blue is usually injected into the fistula at the time of the surgery to delineate the disease, but this is not always successful due to previous infection and scar tissue formation. Surgical approaches and dissection must be modified based upon the relations between the fistula and the facial nerve or skull base.

Simple fistulectomy is possible and sufficient for fistulae that lie in the subcutaneous tissue and do not involve the facial nerve. Four of our cases fell in this category and were successfully treated with simple fistulectomy with no relapse for more than 5 year.

For fistulae that are in close relation to the facial nerve or have previous infections, localization of the facial nerve and/or partial parotidectomy are likely necessary for safe and complete fistula resection. Murthy found modified postauricular and cervical incision helpful in locating the facial nerve and for complete fistula resection in one patient with first branchial fistula. Triglia reported facial nerve dissection and partial parotidectomy in treating 36 of 39 cases of first branchial fistula. He concluded that the surgeon’s understanding of embryology and local anatomy is critical to successful fistula resection. He also concluded that standard parotidectomy incision was beneficial for facial nerve exposure and protection.

The relations between the facial nerve and fistula are complex, and the facial nerve and parotid gland are frequently involved in dissecting the fistula. In 8 of out 12 cases, the course of the fistulae surrounded facial nerve. The “Y” incision around auricular lobule or the standard parotidectomy incision was used in these cases for exposure of the parotid gland and facial nerve. The facial nerve can be approached either from a peripheral branch (e.g., the submandibular branch) or from the main trunk. The former was preferred by the authors, given the difficulties in finding the main trunk in the presence of previous infections and scar tissue formation around the posterior and inferior part of parotid gland.

In rare cases where the skull base is involved, the help from a neurologist may be helpful in facilitating safe and complete fistula removal. Disease resection was incomplete and fistula relapse occurred half a year later in 1 case in this series in
which the fistula extended to the skull base. In this case, preoperative imaging studies failed to accurately demonstrate the entire course of the fistula and the surgeon was therefore under-prepared for the complexity of the procedure. The lesson is to always be prepared for potential complex and difficult disease courses not shown by pre-operative diagnostic studies.

References


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