Surgical management of first branchial cleft anomaly presenting as infected retroauricular mass using a microscopic dissection technique

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Abstract

Purpose: This is a detailed description of the clinical and anatomical presentation of the first branchial cleft anomaly presenting as retroauricular infected mass. Our experience with a microscopic dissection with control of the sinus lumen from within the cyst is also described.

Materials and Methods: Between 2001 and 2008, patients with the final histologic diagnosis of first branchial cleft anomaly in the retroauricular area were managed with a microscopic dissection technique with control of the sinus lumen from within the cyst. Classifications were done in accordance with Work, Olsen, and Chilla. Outcomes measured intervention as a function of disease recurrence and complications including facial nerve function was used.

Result: Eight patients with a mean age of 14.2 years were enrolled, and this included 4 females and 4 males. Four type 1 and 4 type 2 lesions as per the Work’s and Chilla’s classification were found, and there were 5 sinuses, 2 fistulae, and 1 cyst according to Olsen’s classification. All patients presented to the department with acute infection at the time of diagnosis. Five of the 8 patients had previous surgical treatment, 2 of those had up to 3 previous operations. None of the patients were complicated by disease recurrence or had surgical related complications (facial nerve paresis or paralysis, infection, canal stenosis) requiring reoperation with more than 1 year of follow-up.

Conclusions: First branchial cleft anomaly presenting as retroauricular infected mass can be effectively treated by adopting a microscopic dissection technique with control of the sinus lumen from within the cyst.

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1. Introduction

First branchial cleft anomaly (FBCA) represents a rare group of congenital anomaly in the head and neck area. The overall incidence of the lesions is fewer than 10% of all branchial cleft defects [1]. A wide range of clinical manifestations with cervical, parotid, and otologic symptoms can occur with variable presenting loci. The anomalies usually present with acute infection at the initial diagnosis. The leading symptoms are swelling and pain in the region of fistula. Signs of inflammation such as swelling, pain, and erythema are found in more than 70% of the time. In 1 report [2], up to 44% of the patients were complicated by recurrence following failed surgical excision, and 1 patient had up to 7 previous excisions.

In 1972, Work [3] categorized FBCA into type 1 and type 2 anomalies. Work type 1 anomaly usually presents in the very young children as a thin-walled, soft mass protruding in the external auditory canal (EAC) almost causing complete occlusion. The Work type 2 anomaly presents at a somewhat later age as a fistulous opening within the Poncet area [4], where it is bounded superiorly by the EAC, anteriorly by the mental region, laterally by the parotid region, inferiorly by the hyoid bone, and posteriorly by the upper part of the sternocleidomastoid muscle (SCM). Olsen et al [1] proposed another classification and divided the anomaly into cyst: no opening; sinus, 1 opening; and fistula, 2 openings. Chilla and Miehlke [5] distinguish 3 subtypes of FBCA according to their anatomical relationship with the facial nerve (FN): type
1 follows a lateral course, type 2 follows a medial course, and type 3 divides the FN.

Surgical excision of the anomalous epithelial tract is generally regarded as the treatment modality of choice [6]. Misdiagnosis could lead to inadequate surgical resection with the ensuing high recurrent rate. Iatrogenic injury of the FN is also possible [6]. A safe complete resection may require complete exposure of the FN, because the lesions can be variably associated with the nerve [7]. Accurate identification and complete resection of the entire epithelial tract is crucial in the prevention of subsequent recurrence.

In 2005, Baatenburg de Jong [8] proposed an “inside-out technique” in treatment of preauricular sinus in 23 patients and had a satisfactory result. This method highlights a (1) small incision, (2) glistening lining (“inside”), and the outer wall of the tract (“out”) are dissected free of the surrounding tissue with magnification, and (3) A primary closure is accomplished easily because of small incision and subcutaneous dissection. However, we have developed the similar technique of microscopic dissection with control of the sinus lumen from within the cyst in managing preauricular sinus and FBCA since 2001.

The purpose of this retrospect review is to present our experience in the management of 8 cases of infected FBCA presenting in the retroauricular area. The microscopic dissection technique with control of the sinus lumen from within the cyst is described. The merit of this method in treating these lesions is also discussed.

2. Materials and Methods

2.1. Subjects

Eight patients with the final diagnoses of FBCA treated in Chang Gung Memorial Hospital, Linkou, Taiwan from March 2001 to March 2008 were included. The records of these patients were reviewed and summarized in Table 1. This includes sex; age at initial presentation; the side of anomaly; classification according to Work, Olsen, and Chilla; infection status at presentation; inner tract loci; previous history (including surgical intervention and incision and drainage [I&D]); and complication. Diagnoses were mainly made on the basis of the anatomical location and the histopathology of the cyst. Histologic slides were examined to check for the presence of keratinized stratified squamous epithelium, adnexal skin structures, and cartilage. This study was approved by the Institutional Review Board of Chang Gung Memorial Hospital.

2.2. Surgical technique procedure of microscopic dissection with control of the sinus lumen from within the cyst

The routine use of an operating microscope is mandatory. It facilitates a more delicate dissection by enabling the identification and following of the epithelial tracts. An elliptical excision of the tract orifice with the minimal inclusion of the surrounding skin is performed initially. The inflammatory tissue around the infected cyst (retroauricular mass; Fig. 1) superficial to the SCM can be completely excised without the fear of injuring the FN.

The sinus or fistula content is removed immediately after the sinus is opened with sharp scissors. This enables the sinus to be clearly visualized and followed from both the outside (as in classic procedures) and from the inside (Fig. 2). Each

Table 1

<table>
<thead>
<tr>
<th>Sex</th>
<th>Age (y)</th>
<th>Side</th>
<th>Work*</th>
<th>Olsen*</th>
<th>Chilla*</th>
<th>Infection</th>
<th>Inner tract opening</th>
<th>History</th>
<th>Complications</th>
</tr>
</thead>
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<tr>
<td>1 M</td>
<td>10</td>
<td>R</td>
<td>2</td>
<td>Sinus</td>
<td>2</td>
<td>Yes</td>
<td>Blind sac, 1 to junction of cartilaginous and bony EAC and 1 blind sac deep to SCM</td>
<td>1 × surgery</td>
<td>No</td>
</tr>
<tr>
<td>2 F</td>
<td>23</td>
<td>L</td>
<td>2</td>
<td>Sinus</td>
<td>2</td>
<td>Yes</td>
<td>Blind sac deep to SCM</td>
<td>3 × surgery</td>
<td>No</td>
</tr>
<tr>
<td>3 M</td>
<td>28</td>
<td>L</td>
<td>1</td>
<td>cyst</td>
<td>1</td>
<td>Yes</td>
<td>Junction of cartilaginous and bony EAC</td>
<td>1 × I&amp;D</td>
<td>No</td>
</tr>
<tr>
<td>4 M</td>
<td>19</td>
<td>R</td>
<td>2</td>
<td>Sinus</td>
<td>2</td>
<td>Yes</td>
<td>Blind sac deep to SCM</td>
<td>1 × I&amp;D, 1 × surgery</td>
<td>No</td>
</tr>
<tr>
<td>5 F</td>
<td>10</td>
<td>R</td>
<td>1</td>
<td>Fistula</td>
<td>1</td>
<td>Yes</td>
<td>Junction of cartilaginous and bony EAC</td>
<td>3 × surgery</td>
<td>No</td>
</tr>
<tr>
<td>6 M</td>
<td>28</td>
<td>R</td>
<td>1</td>
<td>Sinus</td>
<td>1</td>
<td>Yes</td>
<td>Junction of cartilaginous and bony EAC</td>
<td>2 × I&amp;D</td>
<td>No</td>
</tr>
<tr>
<td>7 F</td>
<td>4</td>
<td>L</td>
<td>2</td>
<td>Sinus</td>
<td>2</td>
<td>Yes</td>
<td>Blind sac 1 to junction of cartilaginous and bony EAC and 1 blind sac deep to SCM</td>
<td>1 × I&amp;D, 1 × surgery</td>
<td>No</td>
</tr>
<tr>
<td>8 F</td>
<td>16</td>
<td>R</td>
<td>1</td>
<td>Fistula</td>
<td>1</td>
<td>Yes</td>
<td>Junction of cartilaginous and bony EAC</td>
<td>2 × I&amp;D</td>
<td>No</td>
</tr>
</tbody>
</table>

M indicates male; F, female; R, right; L, left; I&D, incision and drainage.

* Work, Olsen, and Chilla: classification (see text).
subsequent tract is opened and followed in a similar way until every blind end is identified. Usually, the blind end or the sinus can be found near the junction of cartilaginous and bony portion of the EAC in Work type 1 (Fig. 3) or deeper in Work type 2. Technically, under magnification, the inner glistening cyst lining and the outer wall of the tract can be dissected free of the surrounding infected tissue by scissors or electrocautery under clear view of the inside wall.

Once the sinus tract is found to course near or dive medially to the main trunk or the branches of the FN, the tract dissection to the medial (deep) limit of the lesion is preceded by the identification and dissection of the FN to establish the relationship between the tract and the nerve. The extirpation of deep portion (medial to the FN) of the tract is completed subsequently. The wound bed is checked for remnants of the sinus. After careful hemostasis, the wound is closed in layers with the routine insertion of a low pressure suction drain. The wound is dressed with the sterile skin closure strips.

An artistic description of Work type 2 (patient 1, 2, and 7 in Table 1) is shown in Fig. 5, which shows the infected retro-auricular mass with two main tracts, one of them extends to the junction of the cartilaginous and bony portion of the EAC, and the other ended with a deep-seated blind sac, medial to the main branch of the FN and between the muscle insertion point at the mastoid tip.

3. Results

The records of these patients are shown in Table 1. There were 4 females and 4 males, with a mean age of 14.2 years at presentation. There were 5 lesions on the right side and 3 on
the left side. Four patients underwent preoperative high-resolution computed tomography as a part of the diagnostic workup and to facilitate surgical planning. There were 4 type 1 lesions and 4 type 2 according to the Work’s classification. Three type 2 lesions required the dissection of the FN. There were a total of 5 sinuses, 2 fistulae, and 1 cyst according to Olsen’s classification. By Chilla and Miehlke’s description, half of the tract followed a lateral course and the other half followed a medial course relative to the FN. All the epithelial tracts and the peripheral inflammatory tissues were removed in its entirety in single-stage in all the patients. Neither recurrence nor FN complication was found. No patients required reoperation in all 8 patients with more than 1 year of follow-up.

4. Discussion

There is a higher chance for an otologist than a head and neck surgeon to encounter FBCA presenting in the auricular and retroauricular area. Most of these lesions presenting in the anterior neck and the parotid region were managed by head and neck surgeons in our hospital, and these are not included in this study. In a review article by D’Souza et al [9], only 3 of the 40 sinus tract had an external opening in the retroauricular region. Otologic symptom presents in 24% of the 39 cases reported by Triglia et al [6]. However, only 8 cases of FBCA presented in the retroauricular region in a report by Stokroos and Manni [2] in a series of 18 cases, and most of them were diagnosed and classified as type 1 anomaly by the Work’s classification. Four of the total 8 patients in our study presented as retroauricular infected mass had a final histologic diagnosis of Work type 2 anomaly. This is more rarely seen and can be easily mistaken as type 1 anomaly as demonstrated in Stokroos and Manni’s report [2]. The location of these presenting anomalies is a little posterosuperior to the Poncet area, which is the usual loci for the type 2 FBCA as described by Triglia et al [6] and other researchers [2] and will be less familiar with by surgeons attempting to manage it.

According to our experience, the distal blind end of the epithelial tract in the 4 cases of type 2 lesions were all deeply embedded within or between the muscles bounded anterolaterally by the main trunk of the FN and posteriorly by the medial aspect of mastoid tip. The sinus opening and the tract of the deep-seated portion between the muscles are tightly compressed and can be easily overlooked or obscured by the inflammation during routine exploration without the assistance of magnification or the guiding probe. This may explain for the reasons of incomplete excision and lead to surgical failure as encountered by us and the other reports. It had been reported that the remnants of the lesion were usually encountered more medially from the area previously explored during revision surgery [2].

Unfamiliarity with the disease, limited experience with the required surgical techniques, and insufficient magnification during an operation, all may contribute to the incomplete excision and subsequent recurrence of the branchial cleft anomaly as stated in the previous literature.

Five of 8 of our patients had previous failed surgical attempt, and it is obvious from our experience and others [6,10] that there is a high possibility of recurrence in managing FBCA if the physician is not familiar with or unprepared to manage this disease entity, especially during the acute infected stage.

Whether or not to perform definitive surgical treatment of FBCA in the retroauricular mass during the acute stage is still debatable. Many authors propose I&D of the abscess as an acute initial management and reserve definitive excision only when the infection has completely subsided. It is reported that [10] there is an up to 20% recurrence rate when the definitive excision is performed during the acutely infected period as opposed to a low 3% recurrence rate in uninfected or previously nonsurgically treated cysts [11]. However, repeated I&D or previous incomplete excision may also cause soft tissue scaring that makes future dissection more difficult.
It is controversial in choosing the appropriate time of offering definitive treatment of the infected preauricular sinus. Some authors recommend single-stage definitive excision of the entire epithelial tract including both the cyst and duct at the initial presentation of acute infection with satisfactory results [12]. However, is it also feasible in treating FBCA choosing the same principle as treating preauricular sinus?

Most surgeons agree that FBCA is more complicated compared with that of the preauricular sinus in regards to the anatomical distribution and relationship with adjacent structures with more tract variations described. As a result, surgical management may be more difficult and carries a higher likelihood of disease recurrence and higher risks of injuring the FN compared with the preauricular sinus. It is recommended that a combination of detailed history and examination with appropriate imaging should enable surgeons in reaching an accurate diagnosis. Hence, appropriate referral to senior otologists, patient counseling, consent with the formulation of the desirable definitive surgical approach, and planning form the overall goal of successful management of this particular disease entity. This retrospective study was able to demonstrate that the microscopic dissection with control of the sinus lumen from within the cyst is feasible in the successful management of FBCA, much like managing preauricular fistulae, even at the acute inflammatory stage.

The described surgical technique offers the advantages of creating a clearer operating field in the epithelial tract identification. By directly visualizing the inner lining of the tract helps surgeons in identifying even the smallest tracts in the deepest confined space as in our deep-seated lesions. This is true even when the operation is done during the acutely infected stage. The operating surgeon requires peeling and removing the entire tract wall by separating the outer wall from the peripheral normal tissue bimanually. Even if the cyst wall is ruptured, one can attempt to locate the loci and manage the outpouring sinus wall and granulation as necessary. This method carries the merits of precluding the I&D procedure, and primary closure can be accomplished without difficulty due to the small incision with minimal sacrifice of the peripheral normal tissue in the subcutaneous plane of dissection.

The close proximity between a FBCA and the FN comprises another challenge in the surgical management of this entity. The tract may run deep, superficial, or between the branches of the FN [7]. Awareness of this variability and a readiness to expose and dissect out the nerve are essential when undertaking surgical treatment of these lesions.

In 3 Work type 2 cases in our study, the tract was found to dive deep and medial to the FN with termination in a blind sac adjacent to the styloid process region. The sacs were located in a confined space within the digastric muscles fiber insertion at the medial aspect of mastoid processes. Facial nerve dissection was necessary and performed before proceeding to the final dissection of medial end of the tract as a part of the prophylactic maneuver in protecting the nerve.

In our experience, we did not find it necessary to proceed with the routine identification and dissection of the FN in all cases at the beginning of the procedure as distinct from the recommendations of some reports [6]. There were no patients with complication of FN paresis or paralysis in our study with more than 1 year of follow-up.

It should be stressed that even when the FN is identified and monitored, FN injury is still a well-recognized complication (18% temporary, 1% permanent) in one report [6]. Therefore, adequate patient counseling and consent should be sought at the outpatient department.

Literature indicates that preoperative imaging study [6,13] may provide us with the information of the size and extent of the anomaly along with the locoregional complication of the infective process. Four of our patients underwent preoperative high-resolution computed tomography as a part of the assessment. We found that the relationship of the FN and the deep portion of the tract are difficult to establish preoperatively. Therefore, this further supports that the operating surgeons should always be prepared to expose the FN when necessary and should obtain appropriate informed consent preoperatively.

The diagnosis of FBCA should always be included as a part of the differential diagnosis in the assessment of the retroauricular mass. Other possibilities that must be borne in mind include dermoid cyst, benign inflammatory adenitis, cystic hygroma, lipoma, neurofibroma, hemangioma, sarcoidosis, cystic metastatic carcinoma, lymphoma, and primary parotid tumor.

The result in our study indicates that FBCA presenting as retroauricular mass can be effectively treated, even at the acute infectious stage, by a surgical technique of the microscopic dissection with control of the sinus lumen from within the cyst.

References


