Available online at www.sciencedirect.com
ScienceDirect
British Journal of Oral and Maxillofacial Surgery 50 (2012) 259–263
BRITISH Journal of Oral and Maxillofacial Surgery
www.bjoms.com

Relation between a first branchial cleft anomaly and the facial nerve

Yu-Xing Guo [1], Chuan-Bin Guo *

Department of Oral and Maxillofacial Surgery, School of Stomatology, Peking University, Beijing 100081, People's Republic of China

Accepted 11 March 2011
Available online 22 April 2011

Abstract

Relations between first branchial cleft anomalies and the facial nerve vary. We reviewed 41 patients' medical records and pathological sections to clarify the relation, and found that those on the right side in young patients, which were Work type II and situated low down, were likely to be deep to the facial nerve.

Keywords: First branchial cleft anomalies; Facial nerve; Superficial parotidectomy

Introduction

Anomalies of the first branchial cleft are uncommon and there are few published reports. They account for fewer than 8% of all branchial anomalies.[1] Typically they present as a cyst, sinus, or fistula associated with the external auditory canal, or with a swelling or inflammatory opening in the neck.[2] They are commonly misdiagnosed and are often treated inadequately before being excised completely. Work classified them into two types based on their anatomical and histological features.[3]

A type I lesion presents as a cystic mass and is purely ectodermal in origin, while a type II lesion presents as a cyst, sinus, or fistula, or any combination of ectodermal and mesodermal origin, which may contain skin, adnexial structures, and cartilage.[2,4] Olsen et al. in 1980 proposed a simplified classification into cysts, sinus, and fistulas.[1] It is common for first branchial anomalies to become infected, which causes appreciable morbidity. Excision is generally accepted as the treatment of choice. Correct diagnosis is essential for proper management, while an incorrect diagnosis will often lead to inadequate treatment (incision and drainage or incomplete excision), which results in recurrence.

The purpose of this retrospective review of 41 patients with first branchial cleft anomalies was to identify the characteristic clinical features, method of diagnosis, and management protocol, and assess the relation between the anomalies and the facial nerve.

Patients and methods

Between 1990 and 2010, 41 patients (22 male and 19 female) with anomalies of the first branchial cleft were diagnosed and treated at the department of Oral and Maxillofacial Surgery, Peking University, School of Stomatology. The medical records were complete, and we reviewed the operation notes and histopathological sections to find out how they were diagnosed and managed, the morbidity, and the relation between the first branchial anomaly and the facial nerve.

* Corresponding author. Tel.: +86 10 62179977; fax: +86 10 62173402.
E-mail addresses: gladiater1984@163.com (Y.-X. Guo), guodazuo@sina.com (C.-B. Guo).
[1] Tel.: +86 10 62179977; fax: +86 10 62173402.

0266-4356/$ – see front matter
doi:10.1016/j.bjoms.2011.03.002

Table 1
Clinical signs and anatomical site.

Anatomical site	Clinical signs			Total
	Fistula	Sinus	Cyst	
Periauricular	11	12	0	23
Parotid	0	2	7	9
Cervical	5	4	0	9
Total	16	18	7	41

Results

The mean age at presentation was 14 years (range 0–64), and the mean period for which the lesion had been present was 9 years (range 1 month–40 years). The lesions were on the right side in 18 cases and on the left in 23. Bilateral lesions, or a family history that suggest a hereditary origin, were not recorded.

Twenty-six patients had been previously treated with different antibiotics. Eight patients had had no previous intervention (20%); the remaining 33 had had at least one operation each (mean 2, range 1–7). The "other interventions" included incision, drainage, incomplete excision, and radiotherapy. A 64-year-old man with a 34-year history had had 6 incomplete excisions and 1 course of radiotherapy.

The typical clinical features were a cyst, sinus, or fistula associated with the external auditory canal, and a swelling or inflammatory opening in the neck (Table 1).

Fourteen patients with a fistula or a sinus were examined by fistulography. To confirm the diagnosis, computed tomograms (CT) were obtained in 6 cases and ultrasound scans (US) in 4. Two patients had both fistulograms and CT.

Six cases were misdiagnosed as tumours of the parotid gland. Twenty-nine patients had parotidectomy and dissection of the facial nerve, and the remainder superficial parotidectomy. Twenty-three patients had either a fistula or sinus syringed alternately with methylene blue and normal saline to distinguish the diseased from the normal tissue.

The relation of the anomalies to the facial nerve varied (Table 2). When we reviewed the histopathological sections and used the Work classification, we found 26 cases of type I and 9 of type II. The remaining cases could be included because we did not have enough information.

Table 2
Relation between the lesions and the facial nerve.

Group	Relation to the facial nerve		
	Superficial	Deep	Between branches
Sex			
Male	10	9	3
Female	7	8	4
Side			
Left	12	9	2
Right	5	8	5
Age (years)			
0–9	7	11	5
10–19	4	2	0
20+	8	2	2
Work classification			
I	13	10	3
II	1	5	3
None[a]	3	2	1
Clinical presentation			
Fistula	5	6	5
Sinus	8	9	1
Cyst	4	2	1
Anatomical site			
Periauricular	10	8	5
Parotid	6	2	1
Cervical	1	7	1

[a] Cannot be attributed to any classification.

Case reports

Case 1

A 22-year-old girl was admitted to our unit in November 2009 complaining of swelling in the left submandibular area. She had presented with the same symptoms in 1998, and they had temporarily been relieved with antibiotics. An operation had been attempted in 2004, when she had complained of a fistula track and intermittent drainage from the site roughly every 2 months.

Examination showed a 3 cm scar with a sinus track inferior and posterior to the mandibular angle, which drained yellow fluid on palpation. The remaining ear, nose, and throat examination and general physical examination were within normal limits. CT with fistulography showed that the lesion was close to the left external auditory canal and deep in the parotid gland (Fig. 1).

We exposed the left parotid gland and excised the scarred skin with the fistulous track, together with normal parotid

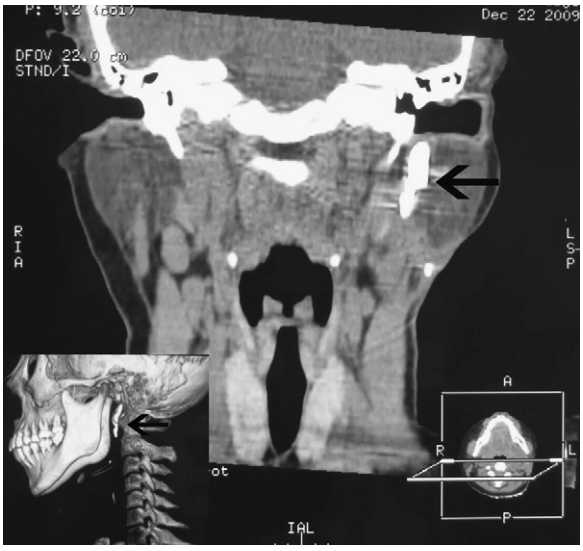

Fig. 1. The computed tomogram and CT three-dimensional reconstruction clearly shows that the lesion is close to the external auditory canal and lies deep to the parotid gland (black arrow).

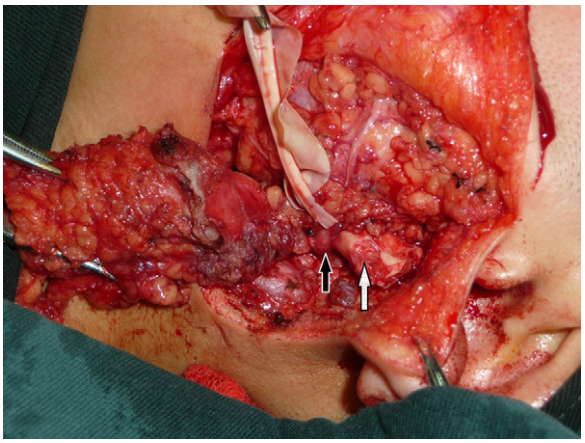

Fig. 2. The lesion (white arrow) lies deep to the facial nerve (black arrow) and in close contact with the left external auditory canal.

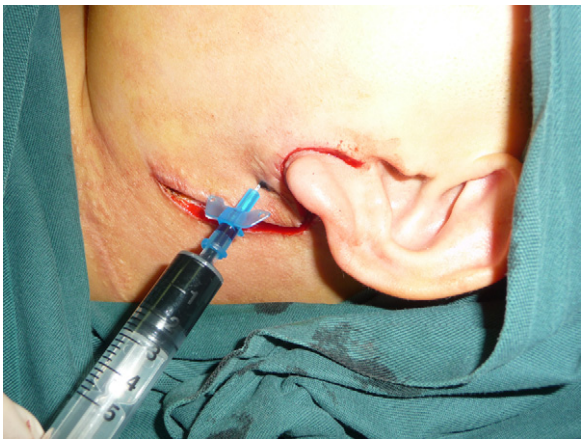

Fig. 3. The fistula syringed with methylene blue.

tissue. The anomaly was enveloped in scar tissue and situated deep to the facial nerve (Fig. 2), which was dissected and was undamaged. There was no connection between this anomaly and the external membranous, bony auditory canal or the middle ear. The incision was sutured with a drain in place, and pressure dressing for 5 days. Postoperatively healing was uneventful and there has been no recurrence to date.

Histological examination showed dense scar tissue, parotid gland tissue, lymph nodes, and cartilage that formed the track of the fistula.

Case 2

A 16-year-old girl was admitted in February 2010 complaining of repeated swelling and infection in the left retroauricular region. She had had 3 operations for this in the past. Since the last operation, she had developed 2 or 3 infections in each year, all of which recovered after treatment with antibiotics. The last infection had been 5 months previously.

On examination a lump about 2 cm × 2 cm with a fistula could be palpated in the left retroauricular region. There were no abnormal secretions from the ears and her general physical examination was within normal limits. CT with fistulography showed that the lesion was not connected to the external auditory canal.

We removed the lesion through a standard parotid gland incision, and before we cut the fistula we syringed it alternately with methylene blue and normal saline to stain the diseased tissue (Fig. 3). After the flap had been reflected, the entire lesion was excised. The facial nerve was not exposed completely during the operation, and the lesion was superficial to the facial nerve. There was no connection with the ear. Healing has been satisfactory with no sign of recurrence.

Histological examination showed a dense scar with chronic inflammation, and the fistula was partly composed of cartilage.

Discussion

In 1923 Frazer described vestigial structures within the neck, which could have been the first description of branchial clefts, and suggested likely sites for their occurrence.[5] Anomalies of the first branchial cleft arise from incomplete closure of the ectodermal portion of the cleft; whether the defect is a fistula, sinus, or cyst depends on the degree of closure.[6]

Nearly all first branchial cleft anomalies are located within a roughly triangular area surrounded by the external auditory canal, the tip of the chin, and the middle of the hyoid bone. The incidence of malformation is higher at the top of the triangle near the external auditory meatus, and in the parotid region, than at the base of the triangle in the vicinity of the hyoid bone.

Correct preoperative diagnosis is difficult. Olsen et al. reported 26 cysts among 38 first branchial anomalies located within the parotid gland, and the preoperative diagnosis in almost all cases was a parotid tumour.[1] Careful physical examination that focuses on the external auditory canal and shows a fistula of the external auditory canal, or an asymptomatic membranous attachment between the floor of the external auditory canal and the tympanic membrane, is more helpful than anatomical or histological classifications, in achieving early diagnosis.[2]

We propose that fistulography should be used for sinuses or fistulas, particularly combined with three-dimensional CT reconstruction. The advantages of CT fistulography and three-dimensional reconstruction are that they directly and accurately assess the course of the fistula, and its position in relation to the external auditory canal. For cysts, the recommended imaging method is B-ultrasound.

The relation between the first branchial cleft anomalies and the facial nerve varies, and can be classified into 3 types: superficial, deep to the facial nerve, or between the branches of the nerve (Fig. 4). It is difficult to predict this relation when making the preoperative diagnosis. After several operations the mixed anatomical layers increase the risk of injuring the facial nerve, which could severely affect the patient's quality

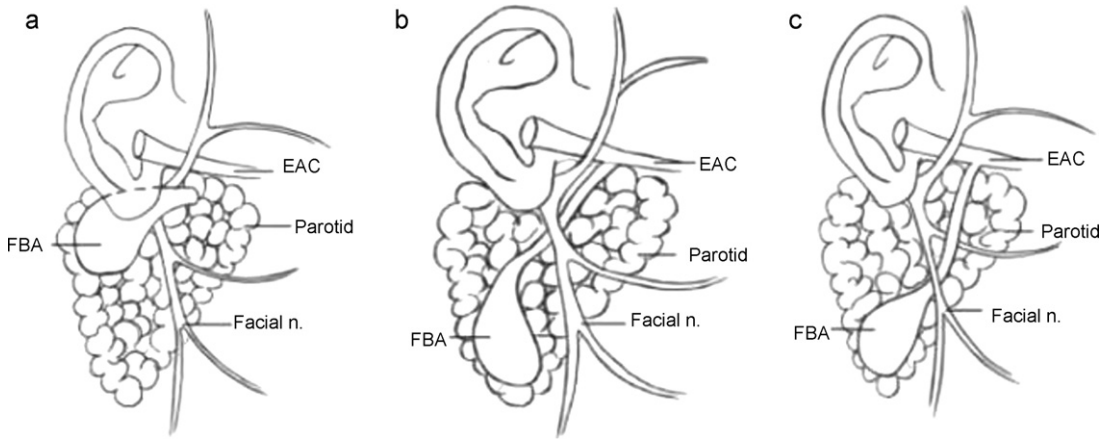

Fig. 4. The track of the fistula lies: (a) superficial to, (b) deep to, and (c) between, branches of the facial nerve. FBA, first branchial anomaly; EAC, external auditory canal.

of life. We reviewed the medical records of our 41 patients during a 20-year period to see if there were any factors that would help us to predict the relation and so protect the facial nerve.

The sex distribution of fistula and sinus was the same (17/17), which is in agreement with the report of Olsen et al., and 5 men had cysts compared with 2 women. Overall the incidence of lesions located deep to the facial nerve was similar in the two sexes. Fistulas and sinuses were more common on the left ($n = 20$) than on the right ($n = 14$), and there were 4 cysts on the right and 3 on the left. Table 2 shows that the lesions on the right were more likely to be deep to the facial nerve.

The younger the patient, the more likely the lesion was to be deep to the facial nerve (Table 2). D'Souza et al. reviewed 55 papers published between 1923 and 2000, a total of 158 cases, and also found that lesions in younger patients were more likely to be deep to the facial nerve.[5] In addition, the facial nerve in an infant is deeper than it is in an adult and there is more risk of it being injured.

Based on the Work classification,[3] we found that type II lesions were more likely to be deep to the facial nerve, but the classification relies on the result of the histopathological section, so not all the lesions can be classified, which limits its clinical value.

The incidence of sinus (6/16) and fistula (9/18) deep to the facial nerve was similar, while cysts were more likely to be superficial (4/7).

From our analysis, we get the impression that first branchial anomalies that are deep to the facial nerve are associated with younger patients; lower lesions; lesions on the right; and Work type II lesions. Because all these cases came from a single hospital and the number is relatively small, the conclusion above may not reflect the overall relation between the lesion and the facial nerve. First branchial anomalies are rare, and the purpose of the initial conclusion is to see if there is anything in that relation that will help to protect the facial nerve.

Some authors have recommended early resection to avoid complications. We recommend that the suitable time for operation is over the age of 4 years, for then the facial nerve has matured and it will be robust enough to cope with the force necessary to raise it and thereby decrease the incidence of facial palsy.[7]

An incision like that used for the excision of a parotid tumour is recommended and enough clinical experience is needed for the meticulous manipulation required to protect the facial nerve.[2,8,9] Those in whom there was a fistula or sinus had it syringed with methylene blue and normal saline alternately to stain the diseased tissue. The staining could also help to distinguish the anomalous track from normal tissue. It is also necessary to excise the involved skin and cartilage of the external auditory canal to prevent recurrence.

According to the relation between the lesion and the facial nerve, the procedure can be either partial excision of the parotid, or superficial parotidectomy. The former applies to lesions superficial to the facial nerve, including fistulas that have stained blue within the region to indicate the amount of tissue to be resected. If the lesion is located deep to, or across, the facial nerve the recommended procedure is superficial parotidectomy. The facial nerve will be completely exposed to traction with a rubber band to ensure the safety of the nerve.

The lower the position of the more deep lesions, the greater the amount of tissue that may need to be removed at superficial parotidectomy. Exploratory surgery should be flexible and respond to the relations between the lesion and the facial nerve. You can always change a superficial parotidectomy into a partial excision of the parotid gland, or vice versa.

References

1. Olsen KD, Maragos NE, Weiland LH. First branchial cleft anomalies. *Laryngoscope* 1980;**90**:423–36.
2. Triglia JM, Nicollas R, Ducroz V, Koltai PJ, Garabedian EN. First branchial cleft anomalies: a study of 39 cases and a review of the literature. *Arch Otolaryngol Head Neck Surg* 1998;**124**:291–5.

3. Work WP. Newer concepts of first branchial cleft defects. *Laryngoscope* 1972;**82**:1581–93.
4. Waldhausen JH. Branchial cleft and arch anomalies in children. *Semin Pediatr Surg* 2006;**15**:64–9.
5. D'Souza AR, Uppal HS, De R, Zeitoun H. Updating concepts of first branchial cleft defects: a literature review. *Int J Pediatr Otorhinolaryngol* 2002;**62**:103–9.
6. Carlson BM. *Human embryology and developmental biology*. St Louis: Mosby; 2004 p. 346.
7. May M, editor. *The facial nerve*. New York: Thieme; 1986. p. 574.
8. Jakubikova J, Stanik R, Stanikova A. Malformations of the first branchial cleft: duplication of the external auditory canal. *Int J Pediatr Otorhinolaryngol* 2005;**69**:255–61.
9. Agaton-Bonilla FC, Gay-Escoda C. Diagnosis and treatment of branchial cleft cysts and fistulae: a retrospective study of 183 patients. *Int J Oral Maxillofac Surg* 1996;**25**: 449–52.