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Clinical Records

First cleft branchial fistula in a child – a modified surgical technique

P. MURTHY, F.R.C.S., P. SHENOY, F.R.C.S., D.L.O., N. A. KHAN, F.R.C.S.

Abstract
Congenital first branchial cleft fistulae, their embryology, anomalies, varied relationships to the facial nerve and surgical techniques for their excision have been well described in the literature. We report a case of a type II first cleft fistula in a three-year-old child which required a modification of the standard surgical approach to achieve safe and complete excision with identification and preservation of the facial nerve.

Key words: Branchial apparatus; Facial nerve

Introduction

Branchial fistulae, cysts and sinuses frequently present to the otolaryngologist either as asymptomatic findings or recurrent infections in the neck, oropharynx or hypopharynx. A brief review of the embryology will assist in understanding the pathogenesis of these conditions.

The primitive pharynx produces five outgrowths known as pharyngeal pouches. Four grooves form on the ectodermal aspect to develop into pharyngeal clefts. The arrangement of these clefts and pouches result in five mesodermal bars which develop into branchial arches. Each cleft, pouch and arch finally give rise to definite structures. Various anomalies of development result in fistulae, sinuses and cysts. These are anatomically related to their embryological source of origin.

Anomalies of the first branchial cleft are rare, accounting for only one per cent of the total anomalies of the branchial system (Farrior and Santini, 1985; Thisted, 1985; May, 1986). These frequently present in childhood with a higher female preponderance (Finn *et al.*, 1987). First branchial cleft fistulae can be divided into two types (Work, 1972). Type I is a duplicated anomaly of the membranous external meatal canal and consists of only ectodermal elements. It starts antero-inferiorly to the pinna, runs parallel to the external ear canal, superficial to the facial nerve and ends blindly towards the middle tympanic region. Type II is a duplicated anomaly of both the membranous ear canal and the auricle and hence contains structures of both ectodermal and mesodermal origin. The tract begins from its caudal opening in the neck below the angle of the mandible and courses upwards piercing the subcutaneous tissue and the substance of the parotid gland. Here it may pass superficially, deeply or between the two divisions of the facial nerve. This variable relationship of the fistula to the facial nerve is postulated to be due to the muscles of facial expression arising from the second branchial arch, crossing the first branchial arch and dragging the fibres of the facial nerve with them (Work and Proctor, 1963; Denecke, 1980).

In view of the intimate relationship of the tract of the type II first branchial fistula with the facial nerve, surgery for excision of these fistulae must closely involve the nerve as well. Hence, appropriate management should consist of exposure of the facial nerve trunk and its branches from the stylomastoid foramen, through the retromandibular fossa to its bifurcation and a little beyond prior to the surgical excision of the entire fistulous tract (Seifert *et al.*, 1986 a,b; May and D'Angelo, 1989). The most widely advocated technique is a preliminary superficial parotidectomy through a standard S-shaped parotid incision with identification and exposure of the facial nerve and its branches (Miller *et al.*, 1984; Lee and Krishnan, 1991). Other methods include stripping of the fistulous tract (Taylor and Bicknell, 1977; Neame, 1983) using wire stilettes, vein strippers and arterial intimal strippers and a turn-inside-out method of the fistula through the ear canal using silk thread (Ohkawa *et al.*, 1990).

Case report

A three-year-old girl presented with a swelling on the left side of her neck of acute onset. A history of recurrent foul smelling discharge from a small opening at the same site as well as from her left external meatus was also obtained. Clinical examination demonstrated an inflammatory globular cystic swelling behind the angle of the mandible at the anterior border of the upper third of the sternomastoid, and also a small opening in the floor of the external auditory meatus at the bony/cartilaginous junction. She was admitted and started on broad spectrum intravenous antibiotics, but in spite of 48 hours of treatment, the cyst became steadily more inflamed needing incision and drainage. Copious cheesy material was obtained. The area settled down satisfactorily and was practically healed by the time of discharge four days later.

Over the following four months, she continued to suffer from recurrent cervical swellings and intermittent discharge from the neck and the fistula in the left external meatus and was admitted for excision of her left type II first branchial cleft fistula.

The operation was commenced with a horizontal cervical incision at the scar site with a view to extend it to a standard S-shaped parotidectomy incision. A probe was passed into the

From the Department of Otolaryngology, Raigmore Hospital NHS Trust, Inverness.
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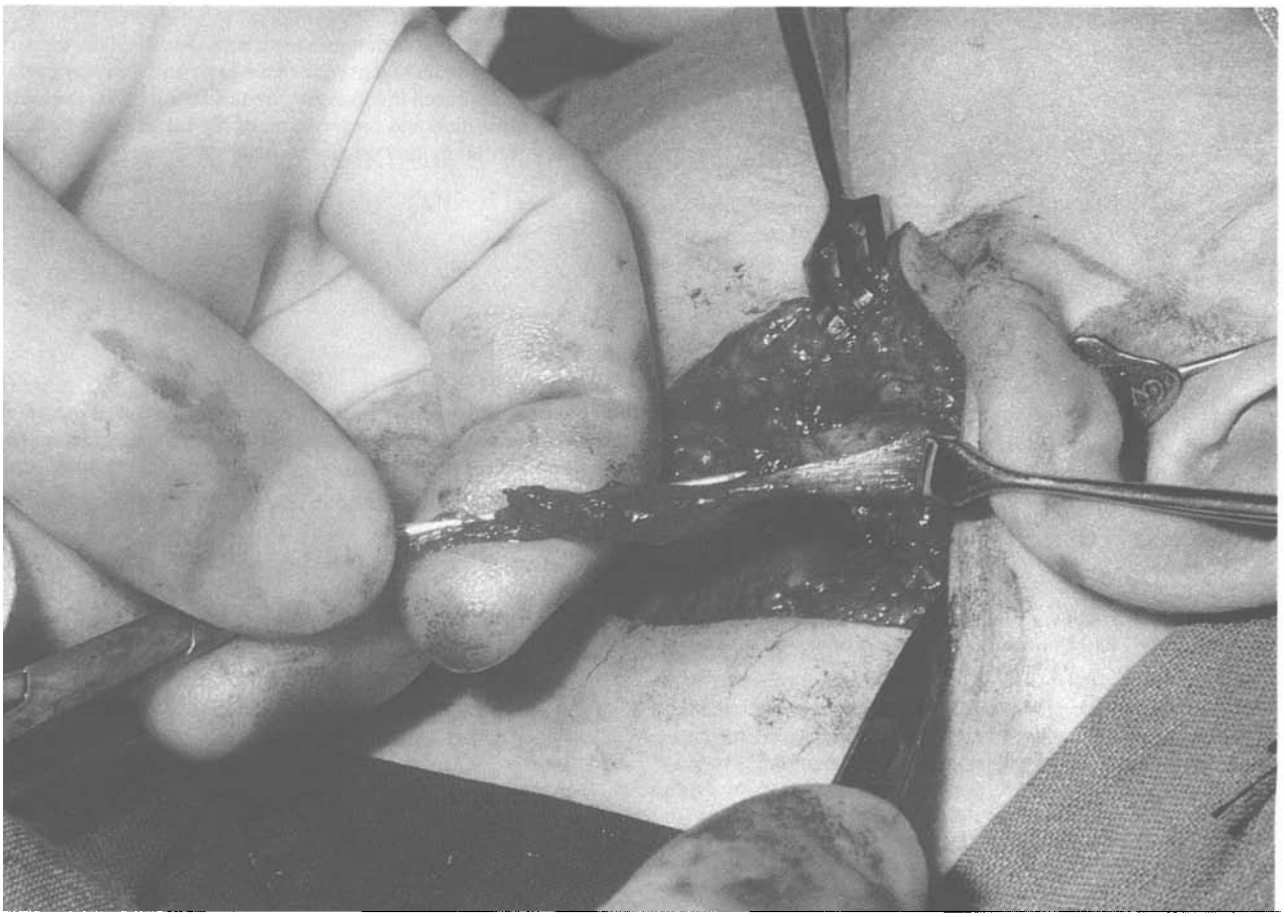

FIG. 1
A probe passed into the fistula through the opening in the ear canal to the wound in the neck.

opening of the fistula in the ear canal down to the level of the wound in the neck (Figure 1). Dissection of the tract was commenced at the lower end and followed towards the external meatus. Approaching this area the tract appeared to course posteriorly and merge into extensive fibrous tissue, a result of previous inflammatory episodes. Identification of the facial nerve proved extremely difficult and the usual landmarks, the surgical pointer and styloid process, were of limited help. To facilitate dissection in order to safeguard the facial nerve the incision was extended postaurally over the mastoid process (Figure 2) instead of anteriorly as originally planned. The trunk of the facial nerve was then easily identified, lying superficial to the fistulous tract. This was confirmed by use of the nerve stimulator which produced a strong contraction of the ipsilateral facial muscles. The tract was completely excised with a sleeve of meatal floor skin and the facial nerve preserved.

Post-operatively the child developed minimal weakness of the facial muscles on the left side as a result of manipulation of the facial nerve during surgery. On close follow-up over the next few months, there was a gradual recovery of the facial movements and six months after the operation, this had completely returned to normal.

Discussion

Surgery for branchial cleft fistulae has been the subject of much controversy for a long time and to date, no single procedure had been universally accepted as ideal. In the case of Type II branchial cleft fistulae, surgery encompasses the extratemporal part of the facial nerve. The variable relationship of the nerve to the fistula and the presence of inflammatory disease and fibrosis make the surgery rather difficult and places the facial nerve in ever greater jeopardy. It is therefore imperative that excision of the fistula must be preceded by identification of the facial nerve.

Additional difficulties in the surgery are encountered in children. The surgical landmarks used for identifying the facial

FIG. 2
Incision extended postaurally over the mastoid process – otological incision.

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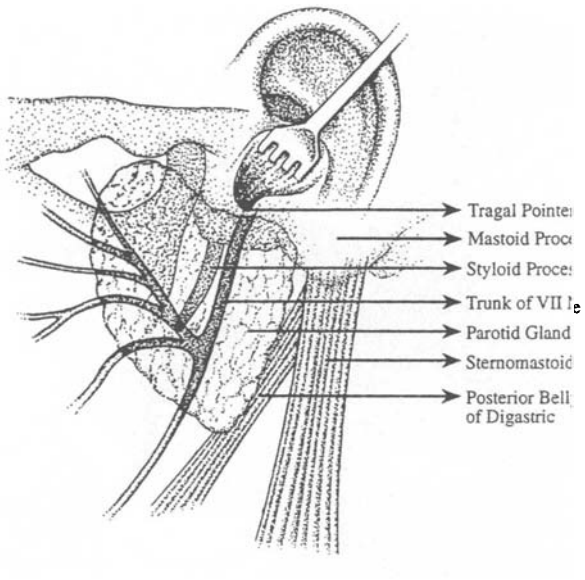

FIG. 3

Surgical anatomy showing the landmarks – tragal pointer and styloid process – for identification of the trunk of the facial nerve in parotid surgery.

nerve near the parotid gland in adults – the tragal pointer and the styloid process (Figure 3) are of limited help. Being more superficial, the facial nerve may appear just after the parotid fascia is separated from the tragal cartilage. Hence dissection to the depth of the 'pointer' could damage the facial nerve (May and D'Angelo, 1989). Using the styloid process as a surgical landmark to find the nerve trunk is also potentially dangerous, as in a third of all patients the styloid process is absent and is impalpable in many others. The styloid process is deeply situated to the nerve and hence the nerve can be injured in a search for this landmark (Seifert *et al.*, 1986 b). The facial nerve may also be damaged if there is accompanying hypoplasia of the parotid gland. This occasional association usually presents with a dimple anterior to the tragus. In this situation the main trunk of the facial nerve and its branches lie subcutaneously without any covering of parotid tissue. Here a standard S-shaped parotidectomy incision could gravely endanger the nerve.

In the case described here, identification of the facial nerve proved difficult due to post-inflammatory fibrosis and the extreme superficial location of the nerve, not uncommon in children. The modified postaural approach enabled us to identify the nerve and safely excise the fistula. We recommend that in similar cases, the facial nerve trunk should be exposed in the retromandibular fossa anterior to the stylomastoid foramen. For this, an 'otological' incision (Figure 2) may be preferred using the landmarks familiar to otologists. Instead of extending the horizontal incision in the neck anterior to the tragus as previously recommended, the incision was carried retroauricularly, taking care to ensure that this is merely skin deep initially as the facial nerve may lie in the subcutaneous plane. The membranous meatus is displaced anteriorly and the suprameatal spine is exposed. The tympanomastoid fissure, which is an excellent landmark for direct exposure of the facial nerve trunk at its exit from the stylomastoid foramen, is exposed lying immediately distal to the suprameatal spine. This is further facilitated by opening up the space between the posterior part of the parotid anteriorly and the sternomastoid and the posterior belly of digastric posteriorly. The nerve trunk usually lies 6–8 mm medially to the end of the tympanomastoid fissure but may lie more superficially in children since it courses laterally immediately after its exit from the stylomastoid foramen. Identification of the facial nerve can be facilitated by the use of a nerve stimulator, particularly in the presence of excessive fibrous tissue. Electrical monitoring of the nerve is also useful in ensuring careful preservation of the trunk and its peripheral branches in difficult cases.

Conclusion

Although several techniques have been described for the surgery of type II branchial fistulae of the first cleft, we recommend a modified approach involving a retroauricular incision to ensure safe identification and preservation of the facial nerve particularly in children and in cases with previous inflammatory episodes.

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Address for correspondence:
Mr P. Murthy, F.R.C.S.,
Department of Otolaryngology,
Raigmore Hospital NHS Trust,
Perth Road,
Inverness IV2 3UJ.