

A unique congenital glossocervical fistula

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Abstract

Lateral cervical fistulae which communicate with the oropharynx are considered to result from incomplete obliteration of the second branchial cleft and pouch. Classically these fistulae have a well-defined pathway through the neck. We present a case, and discuss the aetiology, of a fistula extending from the lateral neck via a previously undescribed course through the neck structures and opening into the posterior aspect of the tongue at the level of the vallate papillae.

Key words: Branchial region; Fistula

Case report

A mildly mentally handicapped 20-year-old man presented with a two-year history of fluid leaking from the left side of his neck after he drank liquids. On examination a small opening was seen on the left side of the neck at the level of the hyoid bone 2 cm from the midline (Figure 1). Fluid was seen to emerge from this opening when the patient drank water. There were no other abnormalities found in the ENT region. A contrast study was performed which demonstrated a dilated tortuous tract extending from the opening in the neck to the level of the pharynx (Figure 2).

It was decided to explore the neck and excise the tract. At operation methylene blue was injected into the opening in the neck and it was seen to emerge from the lateral aspect of the tongue at the level of the vallate papillae. A left transverse skin incision was made to include an island of skin surrounding the opening of the tract. The tract was followed deeply through the platysma and was found to run superficial to the posterior belly of digastric muscle, the stylohyoid ligament and the hypoglossal nerve. It then passed deep to the submandibular gland and into the tongue base, emerging at the level of the vallate papillae. The tract was excised with a surrounding cuff of tongue tissue and the oropharyngeal defect was closed primarily. Post-operative recovery was uneventful and the patient remains well 12 months after surgery.

The gross specimen measured approximately 10 cm in length (Figure 3). It was a squamous lined tract, with relatively little lymphoid tissue. Hair shafts and sebaceous glands were present and there were dilated apocrine glands underlying the squamous lining. Peripheral to the cavity was normal muscle. One end of the specimen was normal skin and other was normal tongue tissue.

Discussion

Congenital lateral cervical fistulae are considered to be derived from the branchial apparatus (Simpson, 1969). Complete fistulous tracts connecting the oropharynx with the external neck are very unusual, those derived from the second arch are the type most commonly reported, with third and fourth branchial arch abnormalities being very rare (Ford *et al.*, 1992).

The vast majority of all branchial disorders are thought to be associated with the second branchial cleft apparatus (Simpson, 1969). Embryologically the second pharyngeal arch enlarges to cover the lower arches forming the cervical sinus deep to it. The



FIG. 1
Discharging cervical fistula.

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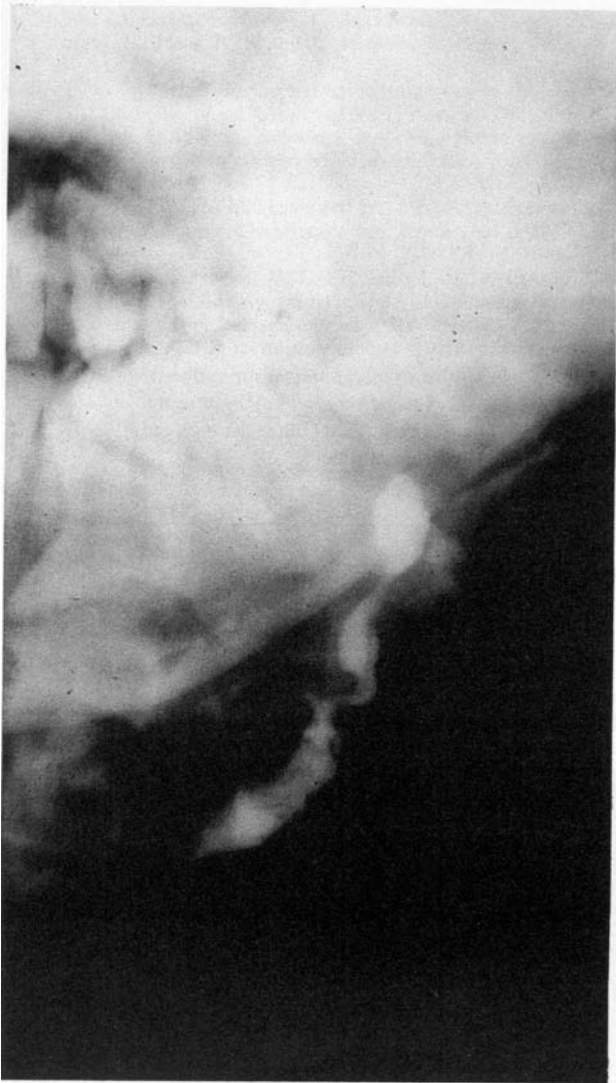


FIG. 2

Contrast study demonstrating a dilated tortuous tract extending from the neck to the oropharynx.

second pouch gives rise to the palatine tonsil with the tonsillar cleft. Normally there is no communication between pharyngeal pouches and clefts and the cervical sinus becomes obliterated. Failure of this obliterative process and the formation of an opening between the pouch and cleft is considered to be responsible

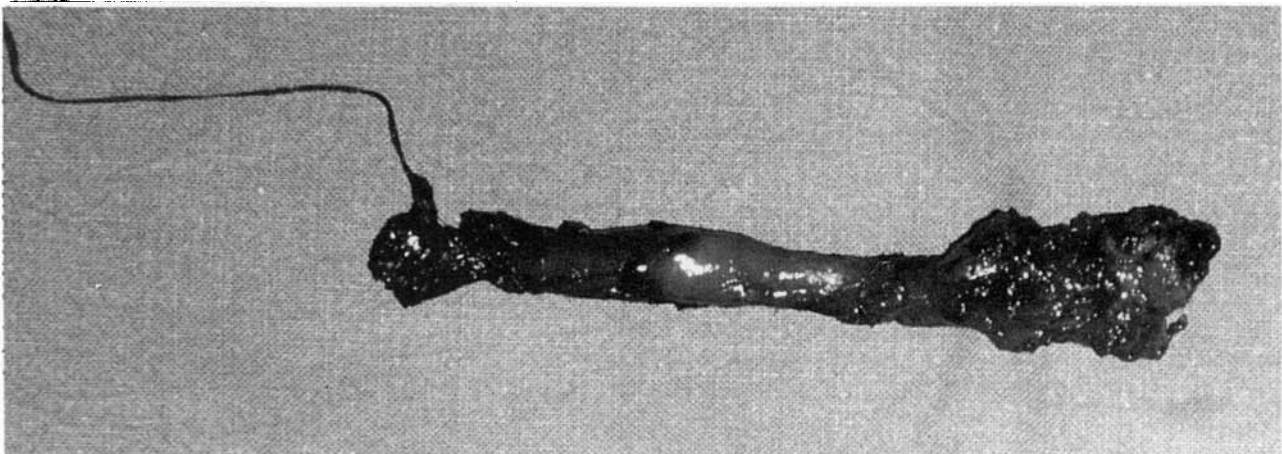


FIG. 3

Excised surgical specimen (tie at skin end).

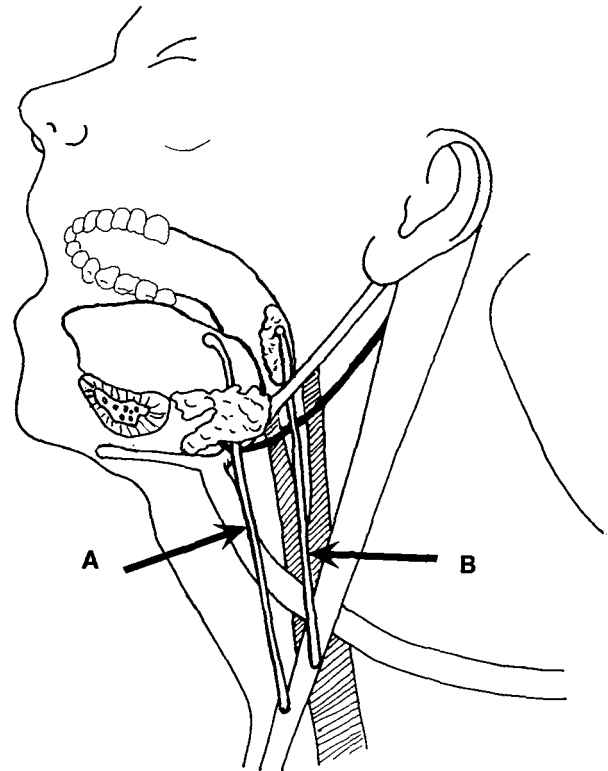


FIG. 4

Diagrammatic representation of the pathways of fistulous tracts through the neck structures: (A) indicates fistula described in report; (B) indicates typical second branchial fistula.

for congenital fistulae between the oropharynx and lateral neck (Rudberg, 1954). These fistulae take a well-defined pathway through the structures of the neck (Gray and Skandalakis, 1972). Commencing at an external opening in the mid to lower neck along the anterior border of the sternocleidomastoid muscle and passing deep to platysma, the tract then courses along the carotid sheath. Above the hyoid it runs medially beneath the stylohyoid muscle and the posterior belly of digastric. It then crosses the hypoglossal and glossopharyngeal nerves before coursing between the internal and external arteries to enter the tonsillar cleft below the stylohyoid ligament. The internal opening is usually on the upper half of the posterior pillar (Davis, 1975) or in the intratonsillar cleft.

The pathway of this fistulous tract compared to that of a second branchial fistula is illustrated diagrammatically in Figure 4. There are a number of unusual features in this case which have

not been described previously. The location of the internal opening in the lateral aspect of the tongue base at the level of vallate papillae is not consistent with that of a typical second branchial arch abnormality nor with fistulae associated with third arch abnormalities (which have their internal opening in the pyriform fossa). Also the pathway of the tract through the neck is anomalous in that it passed superficial to the stylohyoid ligament and posterior belly of digastric, whereas the classically described fistula runs deep to these structures. In addition this fistula did not pass between the internal and external carotid arteries but lay more anterior running underneath the submandibular gland and descending on the carotid sheath. Only the external opening along the anterior border of sternocleidomastoid is in the appropriate location for a typical branchial related fistulae.

Histologically the tract had an outer covering of skeletal muscle and an inner lining of squamous epithelium with skin appendages and collections of lymphoid tissue. These are consistent with the histological features of branchial fistulae described elsewhere (Verbin and Barnes, 1985). The presence of apocrine glands is unusual but probably represent metaplastic changes in the epithelium.

Categorization of this glossocervical fistula is difficult. It most closely resembles a second branchial cleft/pouch fistula, although there are a number of important anatomical differences as detailed in this paper. Other theories of the origin of lateral cervical cysts and fistulae, namely the thymopharyngeal duct theory (Wengelowski, 1912) and the inclusion theories (Bhaskar and Bernier, 1959), are not relevant in this case. Considering that the general location and histological features are consistent with a branchial apparatus origin we feel that this previously undescribed fistulous tract between the oropharynx and the external neck is probably a derivative of the cervical sinus.

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