Cervicoauricular fistula

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Anomalies of the Ist branchial cleft are relatively rare as compared to those of the second. Randall and Royster (1963) indicated the incidence as less than 1% of all branchial cleft anomalies. Lincoln (1965) reviewed the world literature of the past 100 years and found a total of 31 cases and added one of his own. Trail *et al.* (1972) reported three cases out of a series of 56 branchial cleft anomalies. An account of another case was published by Dougall (1974), thereby making up a total of 37. Because of its rarity, and its unusual and interesting features, a case of such malformation is considered worth reporting.

Case report

A 15 year-old-girl was admitted to the Medical College Hospital, Rohtak, with a history of a small swelling on the left side of her neck since childhood. The swelling had occasionally discharged purulent material for the past 3 years. Otoscopic examination revealed a large central defect in the left tympanic membrane. Routine investigations including mastoid radiographs were normal. She was diagnosed as a case of cervico-auricular fistula.

It was decided to explore the sinus and excise the tract. Methylene blue injected into the sinus leaked from a small pit in the floor of the external auditory canal at a depth of about 2 cm. from the surface. A curvilinear incision was made from below the lobule of the left ear to the fistulous opening below and behind the angle of the mandible. The fistulous opening was then threaded with a polythene tube which emerged from the external ear. The wide fistulous tract when dissected was found to pass superficial to the carotid vessels, internal jugular vein, diagastric tendon and hypoglossal nerve (Fig. 1). No effort was made to search for the facial nerve which was probably displaced outwards with the parotid gland. The tract ended in a cartilaginous vestiblue 1 cm. in diameter. The inner end of the tract was divided at the junction of the cartilaginous and bony portions of the external auditory canal.

The excised specimen consisted a 5 cm. long fistulous tract of varying diameter; the inner one third had a cartilaginous feel (Fig. 2 a and b). The cephalic one third was the widest portion of the tract and revealed an ampullary dilatation about 1 cm. in diameter. The lumen of the fistula was 5 mm in diameter in the ampullary region and 2–3 mm. elsewhere. The outer surface of the ampullary region showed seven crescentic depressions (pits) with corresponding elevations inside (Fig 2. a and b).

Sections of the caudal end of the tract revealed stratified squamous epithelium with mild hyperkeratosis. The underlying tissue showed hair follicles and sebaceous and sweat glands. The auricular end contained mature hyaline cartilage with no evidence of lymphoid follicles in the cartilaginous tissue.

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Discussion

Incidence: Congenital anomalies of the first branchial cleft are rare clinical entities which have been reported only sporadically. Most cases are seen during early childhood and young adult life. Females are affected three times as often as males (Trail *et al.*, 1972).

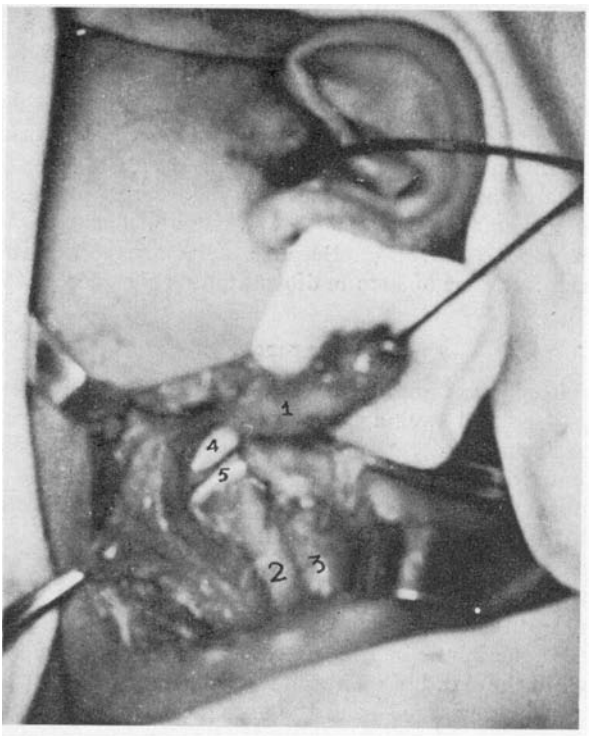


FIG. 1.

Showing fistulous tract (1) threaded with polythene tube emerging from left ear, carotid vessels (2) internal jugalar vein (3) diagastric tendon (4) and hypoglossla nerve (5).

Anatomical considerations

(1) *Relations to the parotid gland:* While there is unanimity of opinion about the tract passing superficial to the carotid vessels and diagastric muscle, its relations to the parotid gland and facial nerve have been reported variably by different workers. The parotid gland was superficial to the tract in the present case and the experience of DeBord (1960) has been similar. Some authors (Leonard *et al.*, 1968) have described the course as superficial to the gland, while Aimi and Takino (1962) traced the course through the gland.

(2) *Facial nerve:* The facial nerve is also reported to have variable relations to the fistulous tract. The nerve may pass deep or superficial to the tract. According to Rankow and Hanford (1953), the time of onset of the anomaly in relation to the change in character of the lateral surface of the foetal pharynx

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may well be an important factor in determining the final course of the VIIth nerve. Whenever interference with the normal process of growth in the region of the branchial cleft is early, then the tract may lie deep to the nerve; conversely, if the time of interference is late, the nerve could be deep to the tract. Exploration and positive identification of the nerve before complete excision has been advocated by Lincoln (1965), because undue stretching of an unrecognized nerve in an anomalous position may lead to paresis of the facial muscles. In the present case no effort was made to demonstrate the actual position of the facial nerve. Presumably it was displaced outwards along with the inferior part of the parotid gland. Gentle retraction probably served as a safeguard against facial palsy.

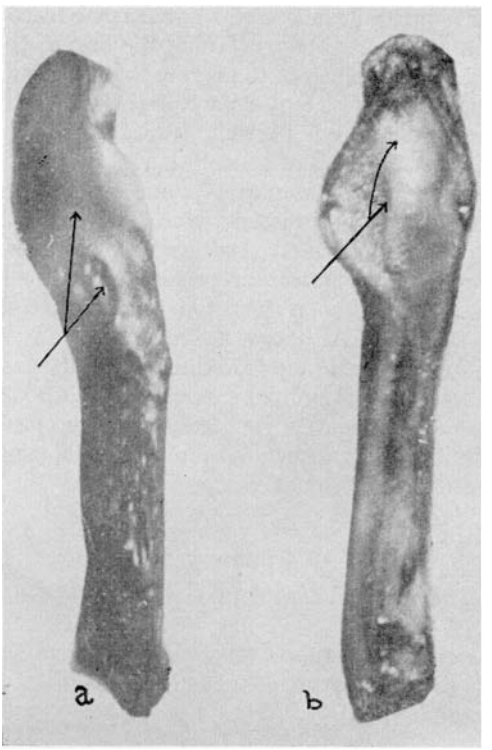


FIG. 2a.
Excised specimen exhibiting ampullary dilatation towards cephalic end bearing pits (marked with arrow).
FIG. 2b.
Fistulous tract laid open, to show elevations and depressions (marked with arrow).

(3) *Location of the tract openings:* The inner opening of the tract in the present case was located at a depth of 3 cm. from the surface. The literature contains references to the inner opening being situated anywhere from the deep part of the external auditory meatus to a superficial position between tragus and antitragus (Frazer, 1923). In the majority of reported cases, including the present

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one, the site of lower end of the fistula is interior to the angle of the mandible and anterior to the middle third of the sternomastoid (Hyndeman and Light, 1929; Shaw *et al.*, 1962; Aimi and Takino, 1962; Lincoln, 1965).

Embryology

The fate of the branchial clefts in the human embryo is extremely vague (Lincoln, 1965). He suggested that a cervicoauricular fistula represents part of the ectoderm of the first cleft which has been pulled dorsally as the ear migrated backwards. This ectoderm may manifest itself as a solid tract over a hollow tube. Lincoln (1965) did not favour the theory of failure of closure and the term was considered to be misleading. He attributed it to necrosis due to a poor blood supply resulting in a breakdown of solid ectodermal tract to form a fistula. Until further evidence is available concerning the fate of the external clefts in the human embryo, it is convenient to assume that a cervicoauricular fistula is a fistula derived from persistence of a part of the first ectodermal cleft, with the coverage of skin in the middle portion of the tract. The presence of a cartilaginous bulb or sheath in 4 cases of Lincoln (1965) were explained as part of a misplaced auricular tubercle or as part of some inductive action of the ectodermal tract on the surrounding mesenchyme. The present case is even more interesting as its cartilaginous sheath displayed 7 pits on its outer surface with corresponding tubercles on the inner side explainable on the basis of duplication of auricular tubercles presenting on the ectodermal surface, which now happens to be the lining of the tract. Although no instance of an associated abnormal condition of the tympanic membrane or middle ear cavity has been reported, the presence of a large defect in the tympanic membrane without a previous history of middle ear disease and a negative roentgenogram for mastoiditis do not seem to exclude the possibility of an associated faulty developmental anomaly of the tympanic membrane in the present case.

Summary

(1) A rare case of cervicoauricular fistula with a review of available literature has been presented.

(2) The cephalic end of a fistulous tract showing elevations and depressions probably represents the duplication of tubercles taking part in the formation of external ear.

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