Simultaneous complete branchial and thyroglossal fistula – a rare presentation

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Abstract The anomalies of branchial cleft and thyroglossal tract accounts for the majority of fistulas in head neck region. A simultaneous presentation of both type of fistula in a same patient is very rare with no such cases reported in Indian literature till date. Here we present a case of simultaneous complete branchial and thyroglossal fistula in a 13-year-old girl with radiologically demonstrable internal opening on fistulogram managed surgically.

Keywords Complete · Branchial fistula · Thyroglossal fistula · Simultaneous

Introduction

The fistulas of head & neck region are among the commonest anomalies encountered by any head neck surgeon. Majority of them results from the anomalies of branchial clefts and anomalies of thyroglossal tract [1, 2, 3]. But a simultaneous presentation of 2nd branchial cleft anomaly along with thyroglossal fistula is a very rare presentation,

more so when the branchial fistula is a complete fistula with clearly demonstrated internal opening in tonsillar fossa. Management of both fistulas is by conventional surgical excision [4, 5].

Case report

A 13-year-old girl presented to the otolaryngology outdoor with complaints of intermittent pain and discharge from a fistula near the midline of the neck slightly to the left since childhood for which she was operated in another state hospital 1 year back. She also complained of intermittent but less frequent pain and discharge from an opening in lower part of right side of neck since childhood (Fig. 1). On examination, the previously operated site was near the midline of the neck 1 cm below the hyoid bone as evident by a scar mark. The opening of the fistula was adherent to the underlying skin with no erythema or tenderness of the adjoining skin. On protrusion of the tongue, the margin of the opening was pulled up. The fistulous opening on the right side of the neck was lying anterior to the anterior margin of the sternomastoid, more or less at the junction of upper 2/3rd and lower 1/3rd. There was no tenderness or erythema of the adjoining skin. On examination, oropharynx was normal with no loco regional lymphadenopathy. On careful examination there was no sign of other congenital anomalies in head neck region or elsewhere. Cranial nerve examination revealed no significant findings.

The patient was subjected to fistulogram apart from the routine investigations. The fistulogram on the fistulous opening on right side of the neck revealed a tract extending up from the opening up to the tonsillar fossa with spillage of dye in the pharynx (Fig. 2). Whereas the fistulogram on the previously operated site could not be performed as the cannula could not be negotiated into the opening due to scarring. But the previous operative notes and investigations confirmed the diagnosis.

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Based on the history, examination and investigations, a diagnosis of simultaneous complete branchial and thyroglossal fistula was made. For thyroglossal fistula, a classical sistrunk's operation was done, which was sadly not done by previous surgeon. For branchial fistula, the fistulous tract was excised using a step ladder approach and the fistula was excised completely up to the tonsillar fossa and the end ligated with silk. Both operations were done in same sitting and the specimen was sent for

Fig. 1 Clinical Photograph showing the fistulous opening in midline of neck with scar mark and an opening at junction of lower and middle third of anterior border of sternomastoid on right side.

Fig. 2 Fistulogram showing a complete branchial fistula with spillage of radio-opaque dye in pharynx.

histopathological examination which further upheld the clinical diagnosis.

Discussion

The fistulas of head & neck region are among the commonest anomalies encountered by any head neck surgeon. Most of them are congenital. The anomalies of branchial cleft accounts for the majority of them, whereas the anomalies of thyroglossal tract account for another major subset [1, 2, 3]. Though individually common, but the simultaneous occurrence of these anomalies in same patient are very rare as seen in our case.

During development, the second branchial arch grows caudally and covers over the third fourth and sixth arch, thus creating a deep pit or sinus covered with ectoderm. The opening of this sinus is normally closed by fusion of its lips so that an ectoderm-lined space is produced. Later normally the cyst is resorbed and disappears but the persistence results in branchial cyst and infection or incision lead to branchial fistula [1, 2, 3]. On the other hand, the thyroglossal fistula results from the persistence of the thyroglossal duct which fail to resorb during the development. Commonest finding is the persistence of the lower part of the tract as the pyramidal lobe of the thyroid [1, 3]. Another common anomaly is the thyroglossal cyst which when gets infected may result in fistula lying in close association with hyoid bone. Developmentally both the fistulas are drastically different and a definite and precise embryological origin of both fistula simultaneously in a same patient is difficult, close development of the thyroglossal fistula between 1st and 2nd branchial arches may be affected in presence of anomalous development of 2nd arch and cleft [2, 3].

The anomalies of second branchial cleft are among one of the commonest congenital anomalies in head neck region. When an external opening is present, the opening is located along the anterior border of the sternocleidomastoid muscle near the junction of the lower- and middle-third portions of the muscle, below the level of the hyoid bone. An internal opening, when present, is located in the tonsillar fossa. When present, a fistula ascends from the lower neck along the carotid sheath and crosses over the hypoglossal nerve and glossopharyngeal nerve. The fistula then passes between the internal and external carotid arteries to end in the tonsillar fossa [1, 2].

Among the fistulas and sinuses, 50% were external-draining sinuses, 39% were complete fistulas, and 11% were internal-draining fistulas. Though complete fistulas are no quite rare but not many convincing, radiologically proven complete fistula is available in literature [2, 3]. A classical fistulogram showing complete length of fistula with internal opening with spillage of dye in the pharynx is quite a rarity in literature, more so when the branchial fistula is associated with thyroglossal fistula.

The standard surgery for second arch anomaly is usually by a stepladder incision originally described by Bailey in 1933. Visualization of the tract at operation may be aided by injecting into the fistula paraffin, methylene-blue dye or quick-hardening polymers. The dissection may be facilitated by prior catheterization of the fistula. Alternatively, fistulas can be excised through the oral route. Stripping of the fistula, first described by Heanley in 1976, requires a correct diagnosis and a complete fistula with both internal and external openings (a complete fistula with external and internal openings is rare). The management of thyroglossal fistula is the classical Sistrunk's operation [2, 4, 5].

The recurrence rate associated with simple excision of a thyroglossal fistula is approximately 50% which may explain the early recurrence of thyroglossal fistula after operation in our case. The recurrence rate can however be reduced drastically with a formal Sistrunk procedure (5%) [1, 3].

Conclusion

Though branchial and thyroglossal fistula are individually very common to any otolaryngologist, their simultaneous

occurrence in a same patient is one very rare finding not reported in any Indian publications so far. This highlights the fact that we should always look for any additional anomalies in any case of head neck sinus or fistula which though very rare is but a reality and little alertness and simple management can prevent unwanted second surgeries.

References

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