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Case Report

A rare case of second branchial fistula with two external openings

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ABSTRACT

Branchial apparatus was first described by Von Baer, but Von Ascheron was the one who described the anomalies in its development. The branchial cyst, sinus and fistula are anomalies of the branchial apparatus which consists of five mesodermal arches separated by invagination of ectoderm (clefts) and endoderm (pouch). Here we present a case of bilobulated second branchial fistula which had both openings over the skin of right anterolateral aspect of the neck. A seven years old girl, presented to ENT OPD, complaining of swelling on right side of the neck associated with discharge from an opening over the swelling since birth. The swelling was painless, progressively increasing in size. Her mother revealed that she had occasionally had a clear discharge from the openings since birth. On examination, dumbbell shaped cystic swelling approx. 5×2 cm in size with two pinhead openings on both the ends, noticed along the anterior border of SCM over the middle third of the neck. Radiological and histopathological investigations confirmed the diagnosis of second branchial fistula. Patient was managed with complete surgical excision. Second branchial anomalies present along the anterior border of sternocleidomastoid. Fistulous tract commonly extends into the pharynx, most commonly in the region of tonsillar fossa. This case report describes a congenital anomaly for which the definitive treatment was surgery. Unlike, other complete second branchial fistula it had two external openings and it was a bilobulated tract, which makes this case a rare variety of branchial fistula

Keywords: Branchial fistula, Fistula, Sinus tract, Congenital, Step ladder incision

INTRODUCTION

Although the branchial apparatus was first described by Von Baer (1827), a mere five years later anomalies in its development were credited with resulting in cervical fistulas (Von Ascheron, 1832). By 1864 the term 'branchial fistula' had been applied (Simpson, 1969).[1]

The branchial cyst, sinus and fistula are anomalies of the branchial apparatus which consists of five mesodermal arches separated by invagination of ectoderm (clefts) and endoderm (pouch).[2]

During embryogenesis, the second arch grows caudally to envelop the third, fourth, and sixth arches; to form the cervical sinus by fusing with the ectoderm caudal to these arches. The ectoderm inside the fused tube disappears, while the edges of cervical sinus fuse. If the ectoderm inside the tube did not disappear this will result in branchial cyst. And if the endoderm breakdown and communicate with skin or the mucosa this will results in branchial fistula.[3,4]

Here we present a case of bilobulated (dumble shaped) second branchial fistula which had both openings over the skin of right anterolateral aspect of the neck.

CASE REPORT

A seven years old girl, presented to ENT OPD, complaining of swelling on right side of the neck

associated with discharge from the openings over the swelling since birth. The swelling was painless, progressively increasing in size. Her mother revealed that she occasionally had clear discharge from the openings since birth. No history of trauma. This was the first time she sought medical care regarding this complaint.

On examination, dumbbell shaped cystic swelling approx. 5x2 cm in size with two pinhead openings on both the ends, noticed along the anterior border of SCM over the middle third of the neck (Figure 1).

No skin changes were noticed in the skin surrounding the openings. Our differential diagnosis was branchial anomalies in the first place, either cyst, sinus, or fistula or lymphatic malformation. Plain MRI neck was done and reported as bilobulated thin-walled cyst like lesion showing fluid contents in the right side of neck at the level of the mandible in the deep fascial and muscle planes, the upper lobulation and lower lobulation are connected to each other with 2 small subcutaneous openings overlying the lobulated components, suggestive of right second branchial cleft cyst fistulas (Figure 2).

After necessary routine investigations, she was planned for right branchial fistula resection under general anaesthesia. Step ladder dissection was performed, circumscribed skin incision was taken around fistulous openings. The fistulous tract was then separated from skin and fascia over it and was completely excised (Figure 3-5). Histopathological examination confirmed the diagnosis. No recurrence of symptoms was seen with 6 months of follow-up.

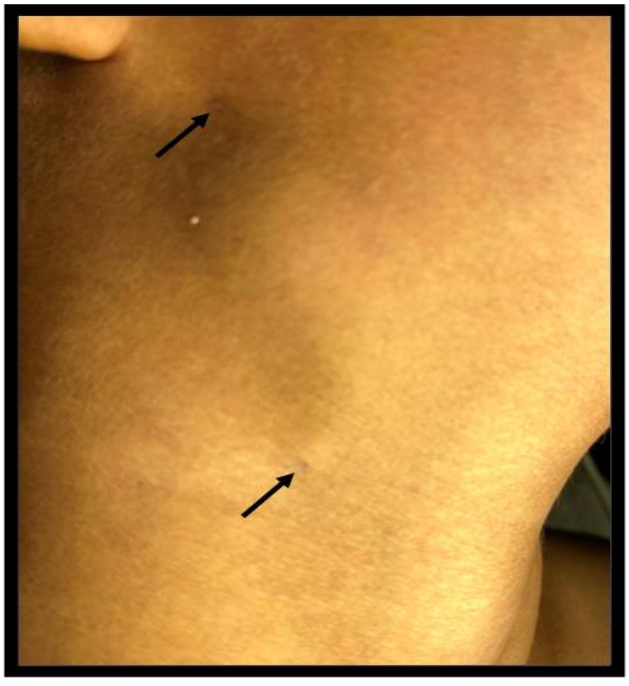

Figure 1: Dumbbell shaped swelling approx., 5×2 cm in size with two pinhead openings on both the ends (arrows).

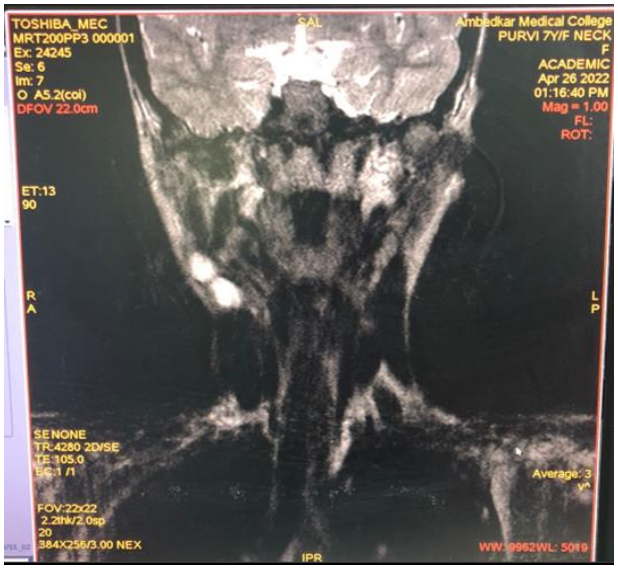

Figure 2: MRI neck showing a bilobulated thin-walled cyst like lesion.

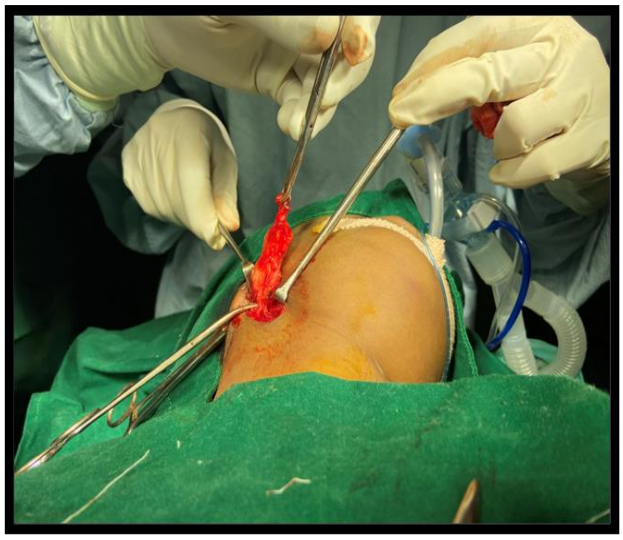

Figure 3: Dissection.

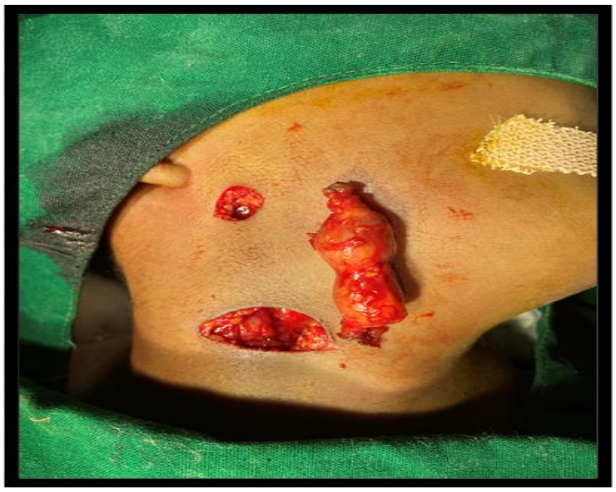

Figure 4: Step ladder approach.

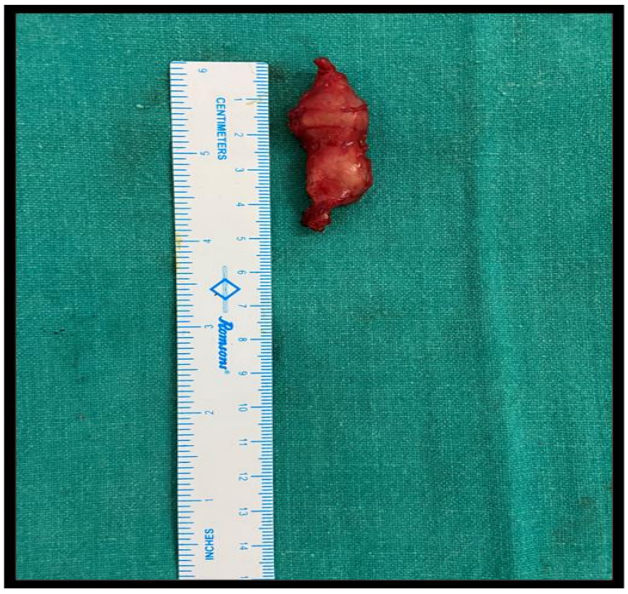

Figure 5: Excised fistula tract.

DISCUSSION

Branchial apparatus consists of three layers: the outer most layer lined by ectoderm; clefts and the inner most layer lined by endoderm; pouches, between them there is a mesodermal lining layer called arches. This was first described by Von Baer.[5]

Anomalies involving branchial apparatus are not fully understood and not clear. There are many theories regarding the origin and classification of these anomalies, yet the most accepted one is the incomplete involution of branchial apparatus during embryogenesis.[6] Branchial anomalies count for 20% of pediatrics congenital neck masses. It may present as cyst which is a remnant of ectoderm and epithelial lining space with no opening into neither the skin nor the mucosa, or it may present as a sinus which is a blind tract into the skin or the mucosa, or fistula a communication between a persistent pouch and cleft to develop.[6-8]

Second branchial anomalies are the commonest, which cover around 95% of all branchial anomalies. A second branchial anomalies are classified into four subtypes according to Bailey's classification: Type 1: Anterior to the sternocleidomastoid muscle and deep to the platysma. Type 2: Deep to the sternocleidomastoid muscle and lateral to the carotid sheath (the most common). Type 3: Lies between the internal and external carotid arteries up to the lateral pharyngeal wall. Type 4: Medial to the carotid sheath. Lying in the pharyngeal mucosal space.

Affected patients commonly present within the first two decades of life. Most of the fistulae are clinically apparent before the age of 5 years. Patients present with persistent or intermittent mucoid or mucopurulent discharge from an opening in the lateral aspect of the neck following an upper respiratory tract infection mostly

Several surgical approaches have been described for the management of a branchial fistula. However, the standard surgery for a second branchial arch fistula is the stepladder approach. It is done through two incisions in the neck that gives exposure of the fistula tract with less tissue dissection. The higher incision must be bigger than the lower to avoid the damage to important neurovascular structures.[9-11]

CONCLUSION

Second branchial anomalies present along the anterior border of sternocleidomastoid in its lower third. Fistulous tract commonly extends into the pharynx entering anywhere from nasopharynx to hypopharynx but most commonly in the region of tonsillar fossa. This case report describes a congenital anomaly for which the definitive treatment was surgery. Unlike, other complete second branchial fistula it had two external openings and it was a bilobulated tract, which makes this case a rare variety of branchial fistula.

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