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Case Report

Bilateral Branchial Sinus: A Case Study

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ABSTRACT

Background: Development of head and neck during fetal life occurs by means of the branchial apparatus which appears during the 4th and 5th week of intra-uterine life. The branchial arches usually involute by the 7th week of intra-uterine life. Failure of the branchial apparatus to involute may lead to various developmental anomalies such as branchial cyst, sinus and fistulas.
Materials and Methods: A 21-year-old girl presented with a history of discharge from right and left sides of lower part of her neck since birth. The patient had bilateral neck swelling over the same site at the time of birth, which burst open spontaneously later in life followed by discharge from the swelling which is sometimes whitish viscous, non foul smelling, painless and sometimes bloody.
Results: The patient was planned to undergo exploration and excision of the fistula. Excision of both sinus tract was done which turned out to be fistulaous tract. Dissection was done with stepladder pattern incision. Fistula was cut and ligated at the upper end of dissection, probing was done with a guide wire. Wound was closed with vicryl 3-0 and skin closed with stapler. The post-operative period was uneventful and the patient recovered completely.
Conclusion: A histopathological examination of the specimen confirmed the presence of a tract lined by pseudostratified columnar epithelium with abundant subepithelial lymphoid tissue.

Key Words: Bilateral branchial sinus, branchial apparatus, branchial, fistula.

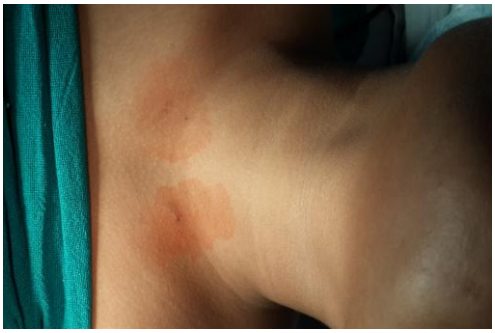
INTRODUCTION

Development of head and neck during fetal life occurs by means of the branchial apparatus which appears during the 4th and 5th week of intra-uterine life. The branchial apparatus consists of six pairs of arches which are mesodermal in origin. Internally the arches are separated by out-pockets of endodermal tissue termed as branchial pouches. Externally the arches are separated by invaginations of ectodermal tissues termed as branchial clefts. The branchial arches usually involute by the 7th week of intra-uterine life. Failure of the branchial apparatus to involute may lead to various developmental anomalies such as branchial cyst, sinus and fistulas. In the embryo the second arch grows in the external direction in a caudal manner to cover the 3rd and 4th arches as well as the 2nd, 3rd and 4th clefts giving rise to a thing ectodermally lined tract called the Cervical sinus of His. The clefts are eventually obliterated as the 2nd arch fuses with the fifth arch to provide a smooth external contour. If however these cleft fail to obliterated they may persist as branchial cysts lined by ectoderm. On the other hand if the 2nd arch fails to fuse with the 5th arch, a thin tract connects the clefts to the skin and this is termed as branchial sinus. If at the same time during the development the membrane between the cleft and pouch ruptures, then the tract communicates

internally and externally and thus aptly termed as branchial fistula. We here report a case of bilateral branchial fistula in a young female. [1-4]

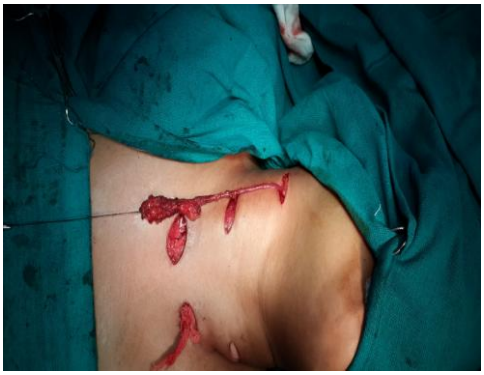
CASE REPORT

A 21-year-old girl presented with a history of discharge from right and left sides of lower part of her neck since birth. The patient had bilateral neck swelling over the same site at the time of birth, which burst open spontaneously later in life followed by discharge from the swelling which is sometimes whitish viscous, non foul smelling, painless and sometimes bloody. This discharge increases after food intake. She also complained of episodes of fever which were associated with increase in the amount of discharge and she received antibiotics for these complaints several times. She was eventually advised surgical treatment. There was no history of recurrent upper respiratory tract infections, dental or jaw infections. There was no history suggestive of Tuberculosis or Malignancy and history of similar swelling on left side at same place in the lower part of neck in her younger brother. General physical examination and systemic examination is within normal limits. Local examination revealed sinus opening bilaterally 2cm away from midline. The sinus opening on left side located just above the sterno-clavicular joint between two heads of left sternocleidomastoid muscle. On the right side a sinus opening was seen on right sternoclavicular joint. Active whitish viscous non foul smelling discharge was present from both sinus openings. Sinus tract was palpable on both sides just lateral to sternocleidomastoid muscle. It moved with deglutition and protrusion of tongue.



Photograph 1: Preoperative bilateral sinus opening

Oral cavity examination was within normal limits. Routine blood investigations and chest X-ray were within normal limits. The patient was planned to undergo exploration and excision of the fistula. Under general anaesthesia, the patient was put in Rose's position. The external opening of the tract was dilated with a probe to delineate the tract. Excision of both sinus tract was done which turned out to be fistulaous tract. Dissection was done with stepladder pattern incision.

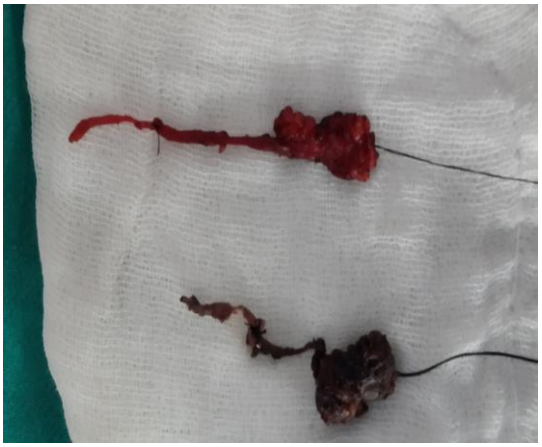


Photograph 2: Intraoperative surgical approach

It was done by deepening the incision and platysma was explored. Dissection extended upto the angle of mandible. Fistulous tract was identified lying superficial to sternocleidomastoid, facial vein, greater auricular nerves and ansa-cervicalis on both sides. Fistula was cut and ligated at the upper end of dissection, probing was done with a guide wire. Tract was extending upto the right tonsillar bed which was identified by laryngoscopy by applying intermittent traction and visualizing the tonsils. A sub-platysmal drain was placed and the sub-cutaneous tissue, platysma and skin were

approximated with 2-0 silk. Wound was closed with vicryl 3-0 and skin closed with stapler.

The patient was extubated, movement of the vocal cords were bilaterally equal. The patient was shifted to post-operative ward in a stable condition. The patient was then put on parenteral antibiotics for 5 days. The post-operative period was uneventful and the patient recovered completely. The patient was discharged on the 5th post-operative day. A histopathological examination of the specimen confirmed the presence of a tract lined by pseudostratified columnar epithelium with abundant subepithelial lymphoid tissue. A final diagnosis of bilateral branchial fistula was made.



Photograph 3: Specimen of both excised branchial tracts

DISCUSSION

Branchial apparatus anomalies are the second most common congenital anomalies of the head and neck region. They usually present during childhood however they may sometimes go ignored to be discovered in adulthood. Most often the branchial fistula arises from the second branchial cleft and usually it is unilateral. Bilateral branchial fistulas are seen in less than 4% cases of branchial fistula. It is not uncommon to find these anomalies to run in families especially in case of bilateral branchial fistula in the form of what is termed as Branchio-oto-renal syndrome or BOR syndrome. BOR syndrome shows autosomal dominant inheritance with presence of ear malformations, pre-auricular pits, renal anomalies and anomalies of the branchial apparatus. BOR syndrome is associated with mutations of the EYA1 gene.

Branchial fistula are often not true fistula since a true internal opening is usually not present is often covered with a thin membrane. Structures arising from the first branchial cleft include the maxilla, mandible, eustachian tube and external auditory canal. The branchial cleft structures are in close relation with the facial nerve and parotid.

In 1977 Work classified remnants of first branchial cleft into two categories i.e. type I and II. Type I are duplication anomalies of the external auditory canal presenting as either fistula or sinus near the post-auricular sulcus or anterior to the tragus. Type II anomalies usually present as a cyst or sinus below the angle of mandible with the tract running superiorly in close association with facial nerve and parotid. [2]

Facial muscles, styloid process, middle ear structures and pinna originate from the second branchial cleft. A branchial fistula most often arises from the second branchial cleft and is typically opens along the anterior border of the sternocleidomastoid, at the junction of the upper and middle third of the muscle. The tract usually passes between the second and third arch structures, i.e. between the internal and external carotid arteries and superficial to the glossopharyngeal nerve. The internal opening can be present anywhere along the nasopharynx but most often opens near the faucial tonsils. The remnants of the second cleft were categorized based on their course by Bailey.

Third branchial cleft anomalies are present in the lower third of the neck and enter the pharynx at the level of the pyriform fossa.

Fourth branchial cleft anomalies open anywhere in the anterior triangle of the neck, the tract runs along the recurrent laryngeal nerve to enter the mediastinum and opens into the upper part of esophagus.

If there is a suspicion of BOR syndrome then the patient should be examined thoroughly and abdominal sonography should be done to rule out renal anomalies. Fistula or sinus tract can be imaged by a fistulogram prior to surgery. MRI provides a non-invasive imaging method to delineate the tract.

The standard treatment of branchial sinus and fistula is total surgical excision with meticulous dissection. The incision is given in a step-ladder pattern. Care should be taken to excise the tract in-toto to avoid recurrence which can be challenging to deal with. An infection of the tract should be dealt with antibiotics and surgery may be delayed until such an event subsides. A relatively newer method includes stripping of the branchial fistula which avoids the extensive dissection. Histological examination of the fistula tract often shows the presence of squamous epithelium or columnar epithelium. Rarely muscle fibers or lymphoid aggregates may also be seen.

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