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COMPLETE SECOND BRANCHIAL ARCH FISTULA: A CASE REPORT

Radiology

KEYWORDS:

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ABSTRACT

The branchial cyst, fistula, and sinuses are the anomalies of the branchial apparatus which consists of five mesodermal arches separated by invaginations of the ectoderm called as clefts. The branchial fistula is not a true fistula as it rarely has two openings. More often even if both ends are patent there is a thin membrane covering the internal opening. Demonstration of a complete branchial fistula on imaging studies is uncommon. We present a case of a complete branchial fistula in a young boy, its clinical presentation, imaging reports and surgical outcome.

CASE REPORT

A 6-year-old male patient presented with history of discharge from the right-hand side of neck on and off since birth. There was no history of trauma or operative intervention. Local examination revealed a pinhead opening along the anterior border of sternocleidomastoid on the right-hand side of lower third of neck. There was no sign of inflammation around the opening.

A fistulogram study was done using iodinated contrast media, which revealed a tract coursing cranially up to the level of right angle of mandible. Patient had gag reflex and could taste the contrast, as the contrast was injected in the caudal fistulous opening, indicating the patent cranial opening in oral cavity or oropharynx (Figure 1)

A contrast enhanced CT fistulogram was done to delineate the exact course of tract before surgery. It revealed a contrast-delineated tract coursing anterior to the right sternocleidomastoid muscle, between internal and external carotid arteries and finally ending at the right tonsillar fossa (Figures 2, 3 and 4). Based on imaging and the clinical history a diagnosis of branchial fistula of second branchial arch was made.

The patient was operated for the fistula with step ladder approach.

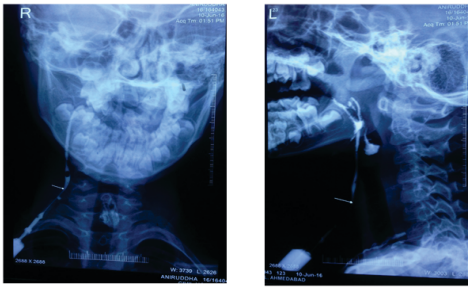

Figure1. Fistulogram, AP and Right Lateral view, showing the contrast delineated fistulous tract (white arrows showing the site of external opening)

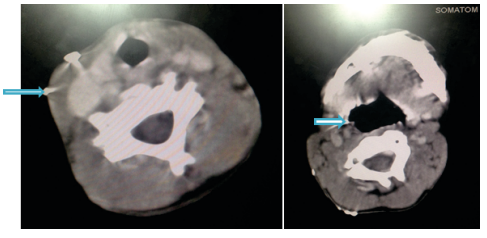

Figure 2.Axial CECT scan of neck showing contrast delineated tract (Arrows showing external and internal opening respectively)

DISCUSSION

The anatomic structures of the face and neck predominantly derive from branchial apparatus, which is complex structure derived from neural crest cells that develops between the second and seventh week of gestation. The branchial apparatus consist of six paired arches separated on their outer surface by five paired ectodermal cleft and on their inner surface by five paired endodermally derived pharyngeal pouches. By the end of fourth week of life, four well defined pairs of arches are visible and the fifth and sixth arches are rudimentary. Each arch is composed of central core of mesoderm and is lined externally by ectoderm and internally by endoderm. The cervical sinus of His is formed after the branchial arches appear by accelerated growth cranially of the first arch and a portion of second arch and caudally by growth of epicardial ridge, which develops from mesoderm lateral to fifth-sixth arch. The branchial apparatus typically disappears between fourth and sixth week of life.[1]

Clinically the second branchial arch anomalies most common presenting features were discharge from the openings, cervical mass, and repeated infection. In addition presence of infection may lead to formation of an abscess and signs of inflammation at the site of opening. [2,3] Because the cysts contain lymphoid tissue, stimulus such as an upper respiratory tract infection can lead to increase in size of cyst.[1] Fistulae are usually diagnosed in infancy/childhood with drainage of secretions or purulent material from an opening at the anterior border of the sternocleidomastoid within the lower third of the neck.[4] With a fistula ostium is often noted at birth at the anterior border of junction of middle and inferior third of sternocleidomastoid muscle. The tract then courses deep to platysma muscle, ascends laterally along the course of carotid sheath, lateral to hypoglossal and glossopharyngeal nerves and then passes between the internal and external carotid arteries before it terminates in the region of palatine tonsillar fossa.[1]

About 90% of all branchial anomalies are related to second branchial apparatus and these anomalies can occur anywhere from the tonsillar fossa to the supraclavicular region of neck. The majority of these anomalies about three fourth are cysts. Cysts are more common between 10 to 40 years of age and fistulas and sinuses usually present before age of 10.[1,3]

Bailey classified the second branchial cleft anomalies into four subtypes based on their location. Types I–III are the most frequently occurring second arch anomalies, with type II being the most common.

Type I Cyst –It lies deep to the platysma muscle and the overlying cervical fascia, anterior to sternocleidomastoid muscle. This type of cyst is felt to represent a remnant

of tract between sinus of His and the skin.

Type II Cyst –Most common and thought to be due to persistence of sinus of His. It is located posterior and lateral to submandibular gland, anterior and medial to sternocleidomastoid muscle and anterior and lateral to carotid space.

Type III Cyst/ Fistula –It is thought to be arising from a dilated pharyngeal pouch and courses medially between internal and external carotid arteries; it can extend upto the lateral wall of pharynx or skull base.

Type IV Cyst –It lies in mucosal space of the pharynx and adjacent to the pharyngeal wall and is thought to arise from a remnant of the pharyngeal pouch.[1]

The diagnosis is most often clinical and radiological investigations are rarely asked for. However, a fistulogram if performed delineates the tract and it is often the commonest investigation available as done in our case. Contrast fistulogram can trace the tract up to the internal opening and is a commonly done preoperative evaluation. Contrast fistulogram can differentiate second and third arch fistulas by demonstrating the internal opening and can obviate the need for further imaging.[5] A complete fistula demonstrable by a fistulogram is uncommon.[3,5] With the available of multislice computed tomography scan a CT fistulogram with reformatted images unambiguously delineates the relation of sinus tract to that of important structures of neck. It also helps in classifying the type of lesion, provides a roadmap for surgeon prior to surgery, and reduces the chance of recurrence.[3]

MRI is most advantageous for Type I first branchial cleft cysts and for parapharyngeal masses that may be second branchial cleft cysts. By the inherent nature of better tissue contrast it provides the relationship of glandular tissue to the mass (e.g. fat planes between the parotid gland and a parapharyngeal mass) and hence acts as a roadmap prior to surgery.[3] It appears hypointense to slightly hyperintense to muscle on T1WI and hyperintense to muscles on T2WI.

Ultrasound is usually initial imaging modality in patients presenting with the neck mass. Ultrasound can delineate the branchial cleft cyst.1 However, complete delineation of fistulous tract is difficult even with the use of high-resolution transducers. If the anomaly is uncomplicated second branchial cleft cysts are typically well-circumscribed, thin-walled and anechoic with evidence of compressibility and posterior acoustic enhancement. They may contain internal echoes compatible with internal debris.[4] When it is infected it can demonstrate a thicker wall with increased internal echogenicity on ultrasound, increased attenuation on CT and hyperintense T1WI on MRI with variable enahancement.[1]

Sometimes there may be a "beak sign" present, which is pathognomonic of Bailey type III second branchial cyst, where the medial aspect of the cyst is compressed and forms a beak as it extends between the internal and external carotid arteries on axial CT or MRI.[1,4]

The treatment of choice for branchial fistula is surgical excision. Several surgical approaches have been described for the management of a branchial fistula. These include a transcervical approach, either by a stepladder approach or through a long incision along the anterior border of sternocliedomastoid and a combined pull through technique. The standard surgery for a second branchial arch fistula is the stepladder approach originally described by Bailey in 1933 with two incisions in the neck that gives exposure of the fistula tract with less tissue dissection. The higher incision should be bigger than the lower one because the fistula tract is deeper in location in the vicinity of important neurovascular structures. The reported incidence of recurrence rate was 3 where only external approach was used.[3,5] This most probably is due to incomplete excision of the fistula tract in the parapharyngeal space. In complete branchial fistula with a probe in situ, the external approach can be combined with intraoral route. No recurrences have been documented with this combined approach. Surgery can be delayed in infants with uncomplicated branchial fistula up to 3 years of age. Infective exacerbations should always be treated before surgery is planned.[3,5]

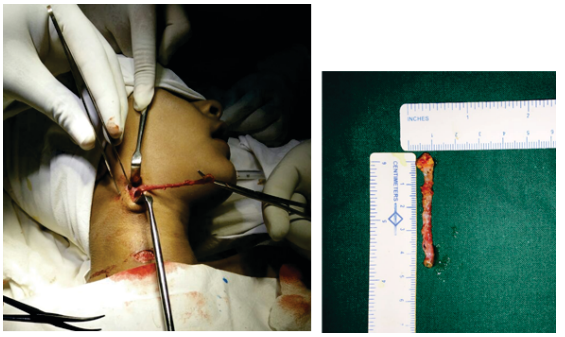

Figure 3. Per-operative specimen of the fistula.

In conclusion, we report a rare complete radiologically demonstrable second brachial arch fistula in a 6 year old male child. The child presented with the external opening at right side of neck with discharge. It was diagnosed by fistulogram and CT fistulogram. The lesion was successfully treated with complete excision using step ladder pattern.

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