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Case report

A Case of Complete Second Arch Branchial Fistula in a 11-Year-Old Child

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Abstract

Introduction: Branchial cleft anomalies are formed due to failure of embryonic structures to obliterate during development. Anomalies in the development of branchial clefts can lead to cysts, external sinuses, internal sinuses, and complete fistulas. The branchial fistula arising from each arch can be identified from the position of the internal and external openings. In majority of cases, the tracts end blindly, leading on to the formation of branchial sinuses. Patients with fistulae often present with mucoid drainage from a lateral neck opening that may become infected over time. Definitive management consists of complete surgical excision of the fistula tract. We report a rare case of complete second arch branchial fistula in a 11 year‑old child, which was successfully treated by excision.

Case Scenario: A 11 years old male presented to Surgery OPD with, chief complaints of opening over right side of neck since childhood with intermittent discharge from the opening. Opening of size 0.1x0.1cm over right side neck at the skin over middle third of anterior border of sternocleidomastoid. Patient was investigated by Conventional X-ray Fistulogram. Patient was further investigated by MRI Fistulogram showing- second branchial cleft cyst with fistula.

Treatment: Patient was operated by brachial fistulectomy by Elliptical skin incision taken around the opening, incision deepened, underlying tract identified and dissected from surrounding structures till just above the level of submandibular gland almost in the area around the tonsillar fossa, the tract ligated at the base and around 6 cm length of tract excised. Postoperatively, patient was kept under surveillance for recurrence.

Conclusion: We reported a rare case of radiologically demonstrable complete second arch branchial fistulae, diagnosed by Fistulogram and treated by complete excision.

Keywords: Branchial Cleft, Branchial Fistula, Fistulectomy, Fistulogram, Stepladder Approach.

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Introduction

Branchial cleft anomalies are formed due to failure of embryonic structures to obliterate during development. These anomalies are the second most common pediatric congenital head and neck masses, accounting for approximately 20% of cases. [1] Anomalies in the development of branchial clefts can lead to cysts, external

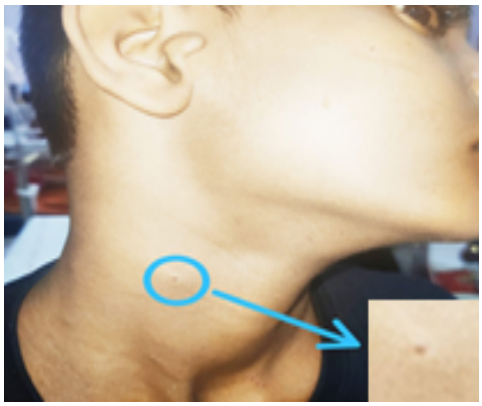
sinuses, internal sinuses, and complete fistulas.[2] Branchial fistulae are formed due to the abnormal persistence of the embryonic second branchial cleft. Branchial fistulae arising from second and third arches are common than from first and fourth arches. The branchial fistula arising from each arch can be identified from the position of the internal and external openings. In majority of cases, the tracts end blindly, leading on to the formation of branchial sinuses. Complete branchial fistula is extremely rare. [3]

Patients with fistulae often present with mucoid drainage from a lateral neck opening that may become infected over time. Given that these patients are at risk for recurrent infection that may lead to abscess formation, definitive management consists of complete surgical excision of the fistula tract. [4] Recurrence has been ascribed to preoperative infection and incomplete excision, though it is likely that recurrence is primarily the result of incomplete excision. [5]

We report a rare case of complete second arch branchial fistula in a 11 year‑old child, which was successfully treated by excision.

Case Scenario:

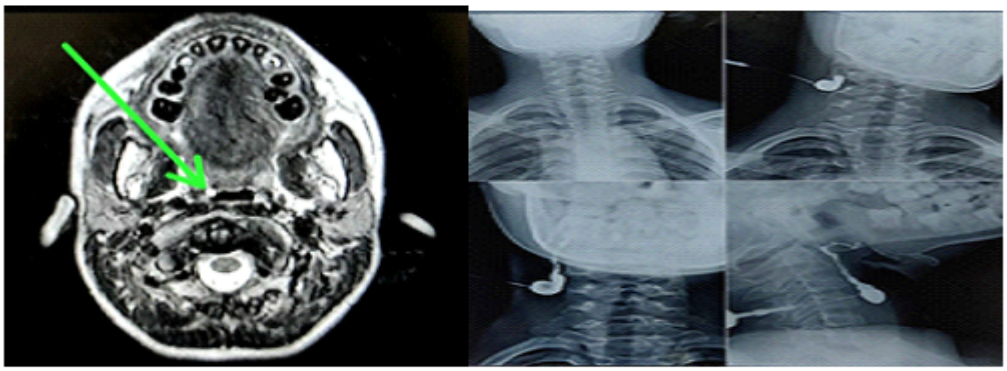
A 11 years old male presented to Surgery OPD with, chief complaints of opening over right side of neck since childhood with intermittent discharge from the opening.



[Image.1]

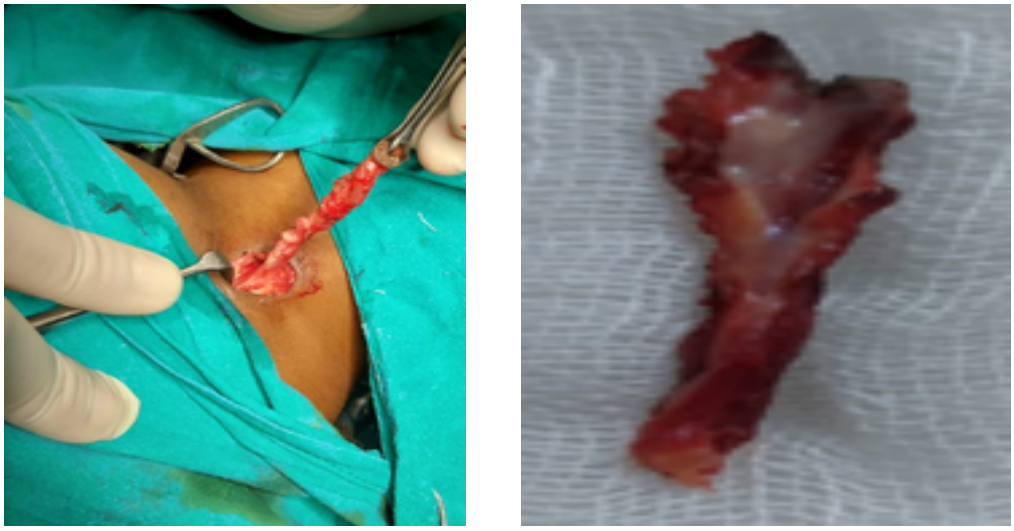
On examination: Opening of size 0.1x0.1cm over right side neck at the skin over middle third of anterior border of sternocleidomastoid with no active discharge [Image.1], per oral examination does not show any evidence of obvious opening or any discharge.

Investigations: Patient was investigated by Conventional X-ray fistulogram. Patient was further investigated by MRI fistulogram showing- second branchial cleft cyst with fistula (type III according to Bailey Classification) [Image.2]



[Image.2]

Treatment: with proper pre-anesthetic evaluation and preparation, after informed and written consent patient was operated by brachial fistulectomy in following steps- Elliptical skin incision taken around the opening, incision deepened, underlying tract identified and dissected from surrounding structures till just above the level of submandibular gland almost in the area around the tonsillar fossa, the tract ligated at the base and around 6 cm length of tract excised. Inner surface of the tract was having mucosal lining with content of tract having mucinous consistency. [Image.3]



[Image.3]

Postoperatively, patient was kept under surveillance for recurrence.

Discussion

The embryonic appearance and differentiation of branchial apparatus occur between the 3rd and 7th week in the human embryo. The mesenchyme gradually grows and obliterates the cleft and pouch in humans. Failure of the second arch tract to obliterate would result in the formation of a branchial sinus and fistula. Patients commonly present in the first two decades of life with intermittent, mucopurulent discharging sinus in the neck. History of recurrent infectious exacerbations and abscess formation may be present. [6] External opening is often situated between the upper two‐thirds and lower one‐third of sternocleidomastoid. Radiologically demonstrable complete branchial fistulae with complete patency from internal to external opening are extremely uncommon in clinical practice. Contrast fistulogram can differentiate second and third arch fistulas by demonstrating the internal opening and can obviate the need for further imaging. Treatment of choice for branchial fistula is complete surgical excision of the fistulous tract. Two surgical methods are commonly used: The stepladder method and the stripping method. Complete excision of the fistula is difficult with external approach alone. Recurrence rate of 3% has been reported with open approach alone, probably due to incomplete surgical excision 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. [7]

Definitive surgical excision for second branchial cleft fistulae is the standard of care. Incomplete excision is considered a major cause of recurrence; thus, some surgeons include ipsilateral tonsillectomy to ensure removal of the fistulous tract. Our data supports findings in current literature that right-sided fistulae are more common.

[8] In a study by Kajosaari et al., 46% of second branchial cleft fistulae excision included ipsilateral tonsillectomy, performed by otolaryngologists. [4]

Fistulae are present at birth but a majority of times a small pinpoint external opening may go unnoticed. The fistula is more commonly seen in males (60%) and 2 to 10% of them can be bilateral.7 When unilateral, 70% of them occur on the right side; 39% are complete fistulae, 50% are external draining sinuses, and 11% have internal opening alone.3 Symptoms include intermittent or continuous mucous discharge or sometimes mucopurulent and recurrent attacks of inflammation following an attack of upper respiratory tract infection. [9]

The diagnosis is most often clinical, and radiological investigations. The fistulography is an effective method of showing the exact anatomy and topography of these fistulae tract in the neck. In our study, fistulogram demonstrated complete fistula in three cases, while in two cases it was demonstrated intraoperatively when the dye was injected. Recently, with the availability of multi slice CT scan, a CT Fistulogram with reformatted images unambiguously delineated the relation of sinus tract to that of important structures of neck. Several surgical approaches have been described for the management of a branchial fistula. [10] 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. [11,12]

Conclusion

In conclusion, we reported a rare case of radiologically demonstrable complete second arch branchial fistulae in a 11 years old male child. The child presented with intermittently discharging sinus in the neck and the diagnosis was confirmed by fistulogram. The lesion was successfully treated by complete excision.

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