Case Report

A rare case of complete second arch branchial fistula in a 7-year-old child

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ABSTRACT

Branchial fistulae are formed due to the abnormal persistence of the embryonic branchial clefts. Complete branchial fistula with internal and external opening is extremely rare. We report a rare case of complete second arch branchial fistulae in a 7-year-old boy, which was confirmed by a fistulogram. The tract was completely excised and the patient was successfully treated.

Key words: Arch, branchial, cleft, complete branchial fistula, fistula, sinus

Introduction

Anomalies in the development of branchial clefts can lead to four unique but closely related lesions, cysts, external sinuses, internal sinuses, and complete fistulas. Complete branchial fistulas are extremely uncommon with only very few reported cases in literature.[1] Demonstration of such complete fistulas by fistulogram is considered rare.[2] The branchial fistula arising from each arch can be identified from the position of the internal and external openings. 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. In majority of cases, the tracts end blindly, leading on to the formation of branchial sinuses. Complete branchial fistula is extremely rare.[3] We report a rare case of complete second arch branchial fistula in a 7-year-old child, which was successfully treated by excision.

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Website: www.njms.in

DOI: 10.4103/0975-5950.111393

Case Report

A 7-year-old boy presented with complaints of watery discharge from an opening in the left side of neck, on and off since 4 years of age. The discharge occurred only during eating or drinking. There was a history of few episodes of swelling at the site of the opening, associated with fever. On examination, there was pinhead-sized opening seen at the junction of middle and lower third of left sternocleidomastoid at its anterior border [Figure 1]. Watery discharge could be seen coming from the opening on making the child to drink water [Video 1]. A clinical diagnosis of branchial fistula was made.

Preoperative pharyngoscopy was normal. A contrast study was done by injecting gastrografin into the external opening, which showed the internal opening at the lateral aspect of pharyngeal wall in the supratonsillar fossa. The patient was able to feel the taste of contrast material in and the esophagus too showed the swallowed contrast, confirming the diagnosis of "complete" branchial fistula [Figure 2].

The child was planned for surgery under general anesthesia. An elliptical incision was made encircling the opening. Subplatysmal flaps were raised. A prolene suture (size 1) was used to probe the fistulous tract. The fistulous tract was traced along carotid sheath where it turned medially to pass between the internal and external carotid arteries. The tract was deep to posterior

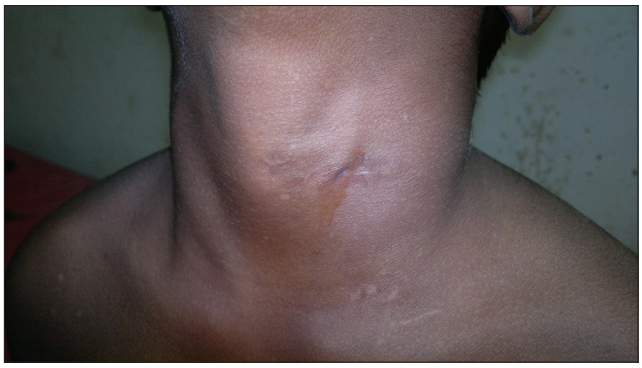

Figure 1: External opening of branchial fistula

belly of digastric opening into the supratonsillar fossa. The fistula was excised completely. Wound was closed without drain. Postoperative recovery was uneventful.

Discussion

The embryonic appearance and differentiation of branchial apparatus occur between the 3rd and 7th week in the human embryo. Five ridges representing branchial arches develop on the ventrolateral surface of the embryonic head. Each ridge has a core of mesenchyme, which is covered externally and internally by ectoderm and endoderm, respectively. Ectodermic clefts separate each adjacent arch externally and endodermic pouches separate them internally. The clefts and pouches move toward each other to form a closing membrane. 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.[4]

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. There is slight female preponderance. Although bilateral branchial fistulas have been documented, unilateral right-sided lesions are commonly seen.[3,4] 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.[5] The tract in second arch fistula extends deep to the platysma, along the carotid sheath, passing between the bifurcation of the carotid arteries after crossing over the hypoglossal and glossopharyngeal nerves and passes below the stylohyoid ligament. It opens internally in the lateral wall of the pharynx region of the tonsillar fossa.[3]

Contrast fistulogram can trace the tract up to the internal opening and is a commonly done preoperative evaluation.

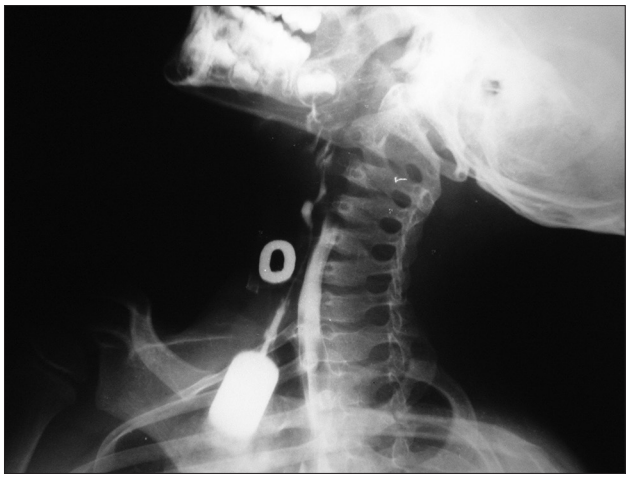

Figure 2: Contrast fistulogram showing internal opening

Contrast fistulogram can differentiate second and third arch fistulas by demonstrating the internal opening and can obviate the need for further imaging.[2] Histology of the sinus tract usually reveals respiratory epithelium with submucosal lymphoid tissue. Rarely, squamous cell lining, mixed cell lining, and branchiogenic carcinoma have also been documented.[1] Tuberculous sinus should be considered possible differential diagnosis.

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.[6] Stepladder approach with two incisions in the neck gives good exposure of the fistulous tract with less tissue dissection. The tract has to be traced till the internal opening and excised completely to prevent recurrence.[7] 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.[1] In complete branchial fistula with a probe *in situ*, the external approach can be combined with intraoral route.[8] 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.

In conclusion, we report a rare case of radiologically demonstrable complete second arch branchial fistulae in a 7-year-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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How to cite this article: Shankar VG, Babu TA, Swami HB. A rare case of complete second arch branchial fistula in a 7-year-old child. Natl J Maxillofac Surg 2012;3:226-8.

Source of Support: Nil. **Conflict of Interest:** None declared.