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Case Report

Surgery Section

Recurrent Branchial Cleft Cyst with Symptomatic Cervical Oesophageal Diverticulum in Adult -An Interesting Presentation of Incomplete Branchial Cleft Cyst Excision

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ABSTRACT

Branchial cysts are congenital cystic lesions of the neck, presenting in childhood. Complete surgical excision is the treatment of choice for these lesions. Recurrence of branchial cysts after incomplete excision is fraught with complications due to the second surgery and the complications of the recurrent cyst per se. Here we present a case of recurrent branchial cyst presenting, after a decade, with a full thickness traction diverticulum of the upper cervical oesophagus necessitating excision of the cyst with the diverticulum. This case report emphasises on the pitfalls of incomplete excision at the index surgery and the long-term complications of recurrence.

Keywords: Re-excision, Second branchial cyst, Traction diverticulum

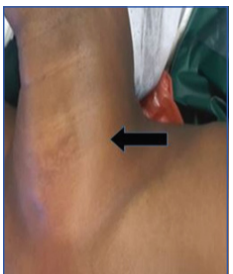
CASE REPORT

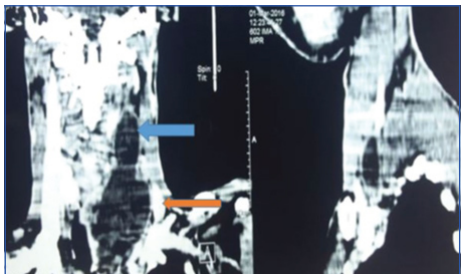
A 25-year-old male patient presented with history of left lateral neck swelling since five years of age and a history of cyst excision done at the age of 13. He was asymptomatic for 10 years until he noticed a recurrent swelling at the same site 18 months back. The swelling was becoming painful and progressively increased in size with intermittent dysphagia for solids and gurgling of liquids for the past month.

On local examination, a single diffuse, fluctuant, non reducible, compressible swelling in the left lateral aspect of the neck was observed [Table/Fig-1]. The lower margin of the swelling was not made out even on hyperextension of the neck. Detailed examination of the oral cavity and pyriform fossa with laryngoscopy showed no evidence of any fistulous opening. Gastroscopy revealed a diverticular opening at 19 cm from the incisors with distal extrinsic oesophageal indentation and narrowing by swelling. CT scan revealed a unilocular thick walled peripherally enhancing cystic lesion of size 13×6 cm in lower cervical/upper thoracic regions-adherent to left oesophagus and trachea medially and the cranial extent involved the left carotid space causing splaying of the internal jugular vein and common carotid artery laterally oesophagogram [Table/Fig-2]. A smooth outpouching at lower cervical and thoracic level with wide opening and distal narrowing of oesophagus was seen [Table/Fig-3]. Fine Needle Aspiration Cytology (FNAC) revealed a paucicellular inflammatory aspirate with no evidence of cholesterol crystals. Though a recurrence of branchial cyst was thought, most likely, other differential diagnosis like foregut cyst, bronchogenic cyst, oesophageal duplication cyst was given due credulence.

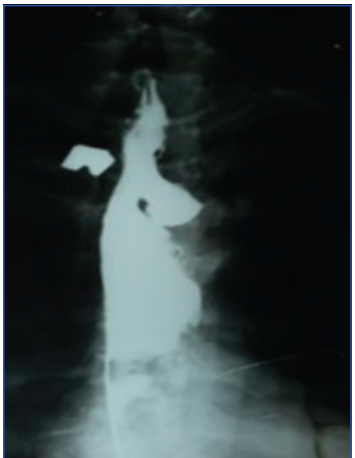
Intraoperatively, a large, loculated cyst was seen medial to carotid and lateral to trachea with no obvious communication between cyst and oesophagus/tonsil. The entire carotid sheath was splayed and oesophageal diverticulum of size 4 cm was seen in posterolateral aspect of oesophagus with blind end adherent to the posterior aspect of the branchial cyst [Table/Fig-4]. The cyst with infected brown fluid was excised leaving behind a small part of the wall adherent to the diverticulum. The broad based oesophageal diverticulum was excised using 60 mm stapler applied to neck of diverticulum under endoscopic guidance.

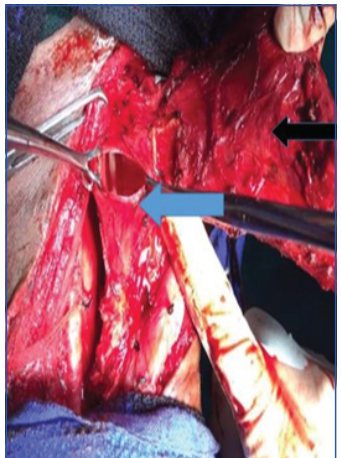
Histology was consistent with branchial cyst which consisted of lining of cyst wall with pseudostratified columnar epithelium



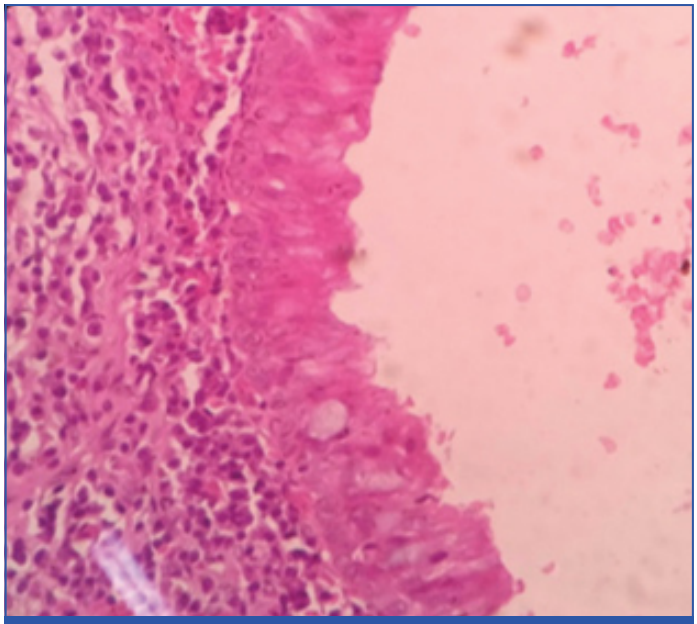


[Table/Fig-1]: Swelling seen in the lateral aspect of the left neck (blue arrow). note: Scar of previous surgery
[Table/Fig-2]: Contrast enhanced computed tomography (CECT) image showing the peripherally enhancing cystic lesion in the left carotid space splaying the carotid artery.





[Table/Fig-3]: Barium study showing wide mouthed left cervical/upper thoracic oesophageal diverticulum.
[Table/Fig-4]: Intraoperative picture showing the cystic lesion (black arrow) adherent to the oesophageal diverticulum (blue arrow) oesophagotmy at the level of the neck of diverticulum reveals the ryles tube passing through the oesophageal lumen.



[Table/Fig-5]: Histopathological examination showing cyst lined by pseudo stratified columnar epithelium with underlying fibrocollagenous tissue showing dense inflammatory infiltrate composed of lymphoid aggregrates and plasma cells with congested blood vessels (H&E, 400X).

[Table/Fig-5] and focal squamous metaplasia and erosions.

The underlying subepithelium showed lymphoplasmacytic infiltrate. The cyst was adherent to true diverticulum which consisted of all its layers including the muscle wall.

It was theorised that the recurrent branchial cyst with adhesions involving the oesophagus had caused a symptomatic cervical traction diverticulum. Postoperative course was uneventful. Patient was on follow up for the past one year with no evidence of recurrence.

DISCUSSION

Branchial cleft cysts develop from the embryological branchial apparatus which develops between second and seventh week of foetal life and are the second most common congenital masses of the neck in children [1].

Persistence of branchial remnants may lead to the formation of cysts, fistulae, or sinuses. The second branchial cleft defect is most common, accounting for more than 90% and are seen in children. Third (2-8%) and fourth branchial cleft (1-2%) defects are uncommon and present later in life with a median age of 5-10 years [2-4]. Most present as left sided swellings (as in our patient) with reports of enlargement with swallowing, filling with air and/or fluid [5,6].

Branchial fistulae and sinuses are diseases of childhood (median age 8.3 years) with branchial cysts mainly occurring in adults (median age 30 years). Most of the branchial cysts (>90%) do not have fistulous communication with tonsil or pyriform fossa as in present case [7]. Second branchial cleft cysts are classified as follows [Table/Fig-6]. This classification has been taken from the article on branchial cleft cyst [8,9].

Type	Classification of branchial cysts
Type I	Most superficial; it lies on the anterior margin of the sternocleidomastoid muscle, deep to the platysma
Type II	Most common; it develops along the anterior margin of the sternocleidomastoid muscle, lateral to the carotid space and posterior to the submandibular gland (the classic location for these cysts)
Type III (as in present case)	Extends medially between the carotid bifurcation and the lateral wall of the pharynx
Type IV	Lies in the pharyngeal mucosal space; lined with columnar epithelium

[Table/Fig-6]: Classification of branchial cleft cysts [8].

Differentiation between third and fourth branchial arch anomalies is fraught with problems as both may have openings in the pyriform fossa and terminate at the skin of the anterior border of sternocleidomastoid muscle [6].

Various methods have been used for treating branchial cleft cysts. Complete surgical excision of the cyst is the treatment of choice for these cysts. Incision and drainage are most commonly used to treat infected branchial cleft cysts, albeit with a high recurrence rate [10]. Open complete surgical removal of fistulous tract in case of branchial fistula is preferred due to low recurrence rate (5% at two years follow up) [2]. In the series by Ford GR et al., the postoperative recurrence rate was 3% [4]. In an another retrospective series by Prasad SC et al., of 34 cases, the incidence of branchial fistula was 20 (58.82%), followed by branchial cyst in 14 (41.17%) cases [11]. The low recurrence rate of 1.2% was believed to be due to better identification of the fistulous tract aided by methylene blue dye, better magnification due to magnification loops or microscope and wide excision of the tract along with the surrounding tissue.

Generally, the aetiology for the increased recurrence might be postulated to be an extension of the cyst through the carotid bifurcation, as might be expected due to suggested origin from second branchial arch remnants [12].

Branchial cysts may present rarely as mediastinal masses, intraoral masses with coexistent retropharyngeal abscess, cranial palsies, ectopic thyroid tissue, and rarely branchial cyst carcinoma, papillary thyroid carcinoma in their walls [13-16]. All suspected branchial cysts must be excised completely to avoid recurrences and complications including like the one mentioned in this rare case report.

This case is reported to underscore the importance of pitfalls of incomplete branchial cyst excision and to the best of our knowledge this is first case report of symptomatic oesophageal diverticulum in the setting of recurrent branchial cyst.

CONCLUSION

Incomplete excision of branchial cyst is a bane associated with increased recurrences and complications including symptomatic oesophageal traction diverticulum as described here. Care should be taken to completely excise these cysts and avoid such complications in the future.

REFERENCES

[1] Wilson DB. Embryonic development of the head and neck: part 2, the branchial region. Head Neck Surg. 1979;2(1):59-66.

[2] Agaton-Bonilla FC, Gay-Escoda C. Diagnosis and treatment of branchial cleft cysts and fistulae: a retrospective study of 183 patients. Int J Oral Maxillofac Surg. 1996;25(6):449-52.

[3] Doi O, Hutson JM, Myers NA, McKelvie PA. Branchial remnants: a review of 58 cases. J Pediatr Surg. 1988;23(9):789-92.

[4] Ford GR, Balakrishnan A, Evans JN, Bailey CM. Branchial cleft and pouch anomalies. J Laryngol Otol. 1992;106(2):137-43.

[5] Mouri N, Muraji T, Nishijima E, Tsugawa C. Reappraisal of lateral cervical cysts in neonates: pyriform sinus cysts as an anatomy based nomenclature. J Pediatr Surg. 1998;33(7):1141-44.

[6] Rosenfeld RM, Biller HF. Fourth branchial pouch sinus: diagnosis and treatment. Otolaryngol Head Neck Surg. 1991;105(1):44-50.

[7] Guldfred LA, Philipsen BB, Siim C. Branchial cleft anomalies: accuracy of pre-operative diagnosis, clinical presentation and management. J Laryngol Otol. 2012;126(6):598-604.

[8] Valentino M, Quiligotti C, Carone L. Branchial cleft cyst. J Ultrasound. 2013;16(1):17-20.

[9] Bailey H. Branchial cysts and other essays on surgical subjects in the faciocervical region. A Book. 1st ed. London, England. Lewis; 1929.

[10] Nicoucar K, Giger R, Jaecklin T, Pope HG, Dulguerov P. Management of

congenital third branchial arch anomalies: a systematic review. Otolaryngol Head Neck Surg. 2010;142(1):21-28.e2.
[11] Prasad SC, Azeez A, Thada ND, Rao P, Bacciu A, Prasad KD. Branchial anomalies: diagnosis and management. Int J Otolaryngol. 2014(2014):237015. pp. 01-09.
[12] Bhaskar SN, Bernier JL. Histogenesis of branchial cysts: a report of 468 cases. Am J Pathol. 1959;35:407-43.
[13] Kotecha V, Muturi A, Ruturi J. Branchial cysts-an unusual cause of a mediastinal mass: a case report. J Med Case Rep. 2015;9:208-10.
[14] Panchbhai AS, Choudhary MS. Branchial cleft cyst at an unusual location: a rare case with a brief review. Dentomaxillofac Radiol. 2012;41:696-702.
[15] Bullock R, Soares DP, James M. An infected branchial cyst complicated by retropharyngeal abscess, cervical osteomyelitis and atlanto-axial subluxation. BMJ Case Reports. 2010;doi:10.1136/bcr.04.2010.2933.
[16] Shin JH, Lee HK, Kim SY, Choi CG, Suh DC. Imaging of parapharyngeal space lesions: focus on the prestyloid compartment. Am J Roentgenol. 2001;177:1465-70.

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