

# Branchial Cleft Cyst Associated with Xanthogranulomatous Inflammation – An Unusual Case

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## Abstract

**Rationale:** Branchial cleft cysts are benign lesions that result from developmental defects arising from primitive branchial arches, cleft, and pouches. Xanthogranulomatous inflammation (XGI) is a mass forming lesion and its association with branchial cleft cyst is rare. **Patient Concerns:** A 23-year-old male presented with a soft, partially mobile, nontender swelling on the left side of submandibular area. His main concern was removal of pathology. **Diagnosis:** Computed tomography scan showed a well-circumscribed rounded cystic lesion suggestive of 2<sup>nd</sup> branchial cleft cyst, which was histopathologically confirmed as branchial cleft cyst with XGI. **Treatment:** Complete surgical excision was done under general anaesthesia and the lesion was removed in toto. **Outcomes:** Patient's postoperative course was uneventful and he was discharged from the hospital on the 4<sup>th</sup> postoperative day. **Take-away Lessons:** Diagnosis of branchial anomalies should always be considered while dealing with lateral neck swellings and XGI associated with branchial cleft cyst should be carefully investigated and treated.

**Keywords:** Branchial cleft cyst, neck mass, xanthogranulomatous inflammation

## INTRODUCTION

Branchial cleft cyst, sometimes called cervical lymphoepithelial cyst, are unilateral, slow-growing benign lesions, commonly present on the lateral aspect of the neck and anterior to sternocleidomastoid muscle. They arise from congenital developmental defects of branchial apparatus between the 2<sup>nd</sup> and 7<sup>th</sup> week of foetal life. 90%–95% of such anomalies are of second branchial cleft and 75% of those were diagnosed in 20–40 years of age.<sup>[1]</sup> Any adult patient with a recent lateral neck mass should always be investigated thoroughly, keeping a suspicious eye for malignancy until proven otherwise.<sup>[2]</sup> Xanthogranulomatous inflammation (XGI) is a mass forming lesion characterised by foamy histiocytes, giant cells, and granuloma formation.<sup>[3-5]</sup> This type of inflammation is rarely described in the head-and-neck area and its association with branchial cleft cyst is unique.

## CASE REPORT

A 23-year-old male was referred to the outpatient unit of the surgery department, with a left side neck swelling of 1-month duration [Figure 1]. There was no history of trauma, tuberculosis, pain, discharge, and no limitation in mouth

opening. Examination revealed a soft, partially mobile, nontender swelling, 5.5 cm × 4.5 cm in size, on the left side of submandibular area with normal overlying skin. No cervical lymphadenopathy was found.

Fine-needle aspiration cytology (FNAC) revealed pus-like fluid mixed with blood showed numerous anucleated squamous cells along with scattered giant cells and few inflammatory cells indicating a benign cystic lesion. Computed tomography (CT) scan showed a well-circumscribed rounded cystic lesion, measuring about 3.2 cm × 2.6 cm × 3.1 cm with peripherally enhancing thin wall in the left submandibular region posterior to the submandibular gland and anterior to the sternocleidomastoid muscle. There was no sign of infiltration into the nearby

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structures. Both lobes of thyroid gland, carotid, and jugular vessels appeared normal. There was no significant enlarged cervical lymph node and no obvious mass lesions in the oral cavity, oropharynx, and larynx. Differential diagnosis included branchial cleft cyst, tuberculosis, salivary gland neoplasm, neurogenic neoplasm, and metastatic lymph node enlargement.

The imaging features and location were suggestive of 2<sup>nd</sup> branchial cleft cyst [Figure 2].

Complete surgical excision was done under general anaesthesia using the left transverse cervical approach. Incision was followed by exposure of platysma with careful dissection and the lesion was found separate from the left submandibular gland and removed in toto [Figure 3].

The lesion was sent for histopathological examination. Macroscopically, specimen was a round unilocular cyst measuring about 4.8 cm × 4.2 cm × 3.0 cm. The cut surface showed a cystic mass filled with purulent material and multiple grayish-yellow nodules were present. Microscopy showed cyst wall with a stroma of lymphoid type, having germinal center in a few areas, covered by stratified squamous epithelium with reactive changes. Cystic lining revealed granulomatous features with foamy histiocytes, giant cells (Touton type) and lymphocytic infiltrate [Figure 4]. Our diagnosis was consistent with a branchial cleft cyst with XGI.

Postoperative course was uneventful and the patient was discharged from the hospital on 4<sup>th</sup> postoperative day and has been in regular follow-up for 9 months and doing well.

## DISCUSSION

Anomalies of branchial arches represent around 20% of cervical masses, present as cysts, sinus, and fistulas. Precise location and course of these anomalies depend on specific branchial cleft and pouch from which they are derived. Second branchial arch anomalies are most common and account for more than 90% of cases.<sup>[1,6]</sup> Multiple theories of origin have been proposed and four main theories

are- (1) incomplete obliteration of branchial cleft, arches, and pouches, (2) persistence of the vestiges of precervical sinuses, (3) branchial ductal origin, and (4) cystic lymph node origin. They are bilateral in 2%–10% of cases but generally unilateral, typically present on the lateral aspect of the neck and commonly seen in young adults.<sup>[1,7]</sup> Any new mass presenting on the lateral side of the neck should be considered malignant until proved otherwise and differential diagnosis should include metastatic lymphadenopathy, tuberculosis, and lymphoma.<sup>[2,7]</sup>

The second branchial cleft cyst is most common and clinically presents as a nontender, compressible swelling on the anterior border of sternocleidomastoid muscle at the junction of its middle and lower thirds.<sup>[1,7,8]</sup>

Proctor (1955) revised Bailey’s classification of second branchial cleft cyst into four subtypes.<sup>[7]</sup>

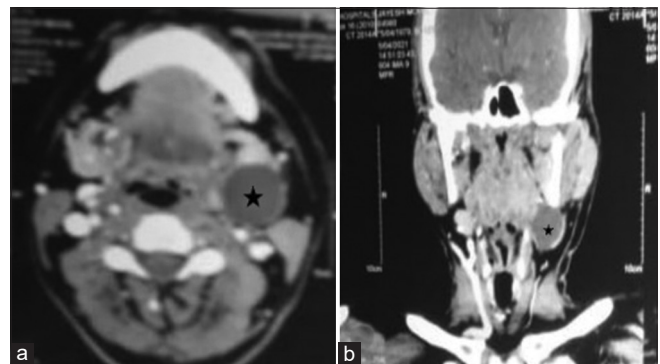
1. Type 1– anterior to sternocleidomastoid and deep to the platysma
2. Type 2 – the most common type, adjacent to internal carotid artery and adherent to the internal jugular vein
3. Type 3 – extends between external and internal carotid arteries
4. Type 4 – adherent to the lateral pharyngeal wall and may extend superiorly to skull base.

Radiological findings, ultrasonography, CT-scan, and magnetic resonance imaging (MRI) are important aids in diagnosis of branchial cyst and help define the exact size of the lesion and topographic relation to adjacent structures. Ultrasonography is suggestive of branchial cleft cyst with well-defined border along with hypoechoic or anechoic lesions without intralesional septation. CT scan usually presented with a thin wall, well-bordered, and uniformly hypodense cystic mass. MRI may vary in branchial cleft cyst between hypointense and hyperintense depending on the protein content of cysts.<sup>[9]</sup> Beware of second branchial arch cyst diagnosis when using computerised tomography, MRI, and even sonography of the neck for diagnosis.<sup>[2]</sup>

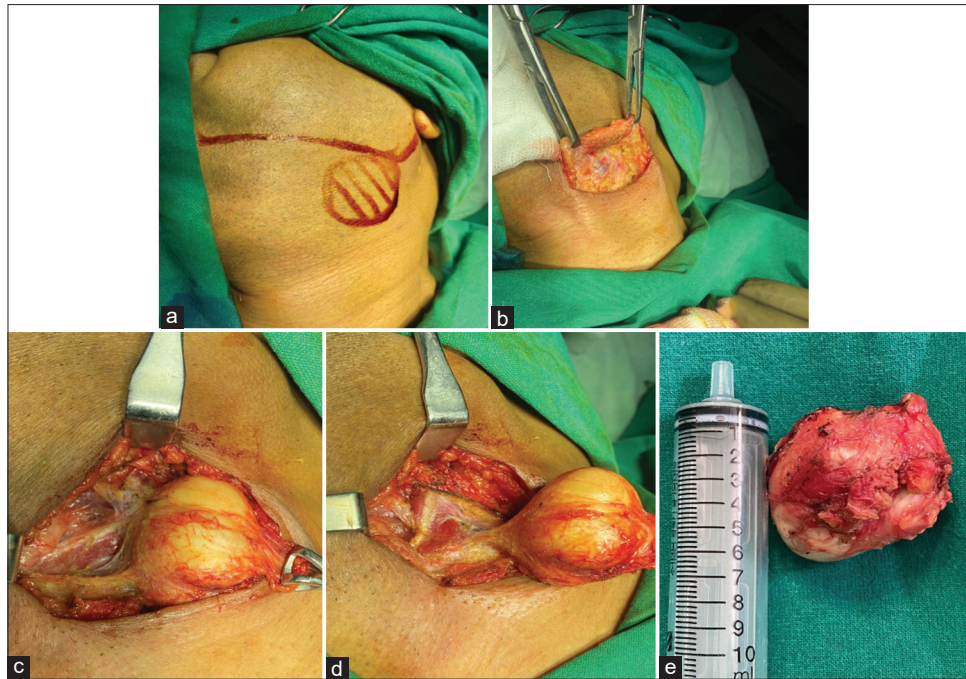
In our case, CT scan showed a well-circumscribed rounded cystic lesion, with a peripherally enhancing thin wall posterior to



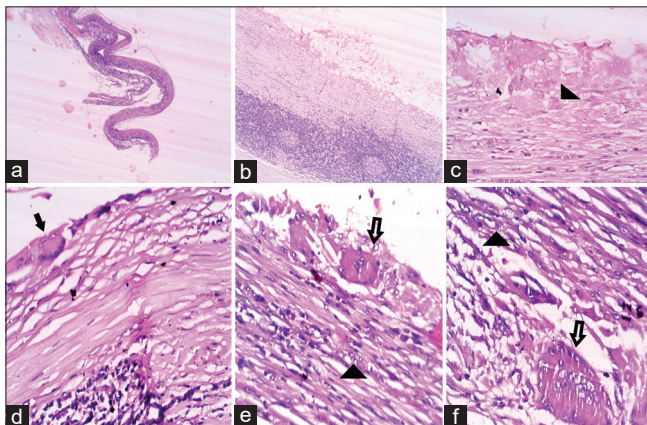
**Figure 1:** Clinical picture



**Figure 2:** Computed tomography scan. (a) Axial view, (b) coronal view showing the cystic area (\*)



**Figure 3:** Operative picture in sequence (a --> e) (a) anatomical surface markings (b) sub-mandibular incision given (c) swelling exposed and dissection done (d) specimen retrieved in toto (e) gross specimen



**Figure 4:** Histopathological picture. (a) Cystic lining with lymphoid stroma (H and E, ×4), (b) Xanthogranulomatous inflammation and cystic wall showing germinal center, (×10) (c) Cystic wall with foamy histiocytes (▼), giant cell and lymphoid stroma (×10), (d-f) Shows granuloma formation with Touton type giant cells (→) and foamy histiocytes (▼) (×40)

the submandibular gland and anterior to the sternocleidomastoid muscle suggestive of second branchial cleft cyst.

FNAC also is a very useful diagnostic tool for surgical intervention. In the study of 18 patients, FNAC was performed in 15 patients and was 100% accurate in diagnosing the benign and cystic lesions. Diagnostic criteria for branchial cleft cyst are anucleate keratinised cells, squamous epithelial cells of variable maturity in a background of amorphous debris along with the accurate anatomic site.<sup>[10]</sup> In our case also, FNAC was showing cystic benign lesion.

Histopathologically, diagnosis is confirmed by stratified squamous epithelial lining, sometimes pseudostratified, ciliated, columnar, and ulcerated. Connective tissue wall consists of abundant lymphoid tissue and may have germinal centre.<sup>[1,3]</sup> XGI is a mass forming lesion characterised by foamy histiocytes, lymphocytes, and Touton type giant cells. It is more common in the kidney and gallbladder.<sup>[4]</sup> Very few cases are described in the head and neck and uniquely restricted to some cystic lesions and masses in the neck including Rathke’s cleft cyst in pituitary gland, colloid cyst in 3<sup>rd</sup> ventricle, branchial cleft cyst, and thyroglossal duct cyst.<sup>[3-5]</sup> Our case is the second case of branchial cleft cyst with XGI. Several theories have been proposed for the association of XGI with cystic lesions in head and neck but actual relationship is still largely unknown. Many hypotheses have been proposed including immunological disorders, defective lipid transport, reaction to a low virulence-specific infection and obstruction. It has been noted that FNAC and rupture of cyst may be associated with XGI.<sup>[4,5]</sup>

Adjacent tissue and organ infiltration is quite frequent in this type of inflammation which has been identified in many regions such as the pancreas, testis, kidney, and gallbladder.<sup>[3]</sup> Distinction between branchial cleft cyst with XGI and other XGIs should be made by identification of cystic underlying disease and clinical, radiographical, and histopathological findings.

## CONCLUSION

Diagnosis of branchial anomalies should always be considered while dealing with lateral neck swellings. XGI associated



with branchial cleft cyst should be carefully investigated. The diagnosis of malignancy should always be a consideration; hence appropriate workup should be done preoperatively. Preoperative diagnosis with FNAC and radiological findings are good diagnostic tools to determine the cystic nature of swellings in anatomic proximity to vital structures and help in future surgical planning and reaching a final diagnosis. Surgical excision is the gold standard for the treatment of branchial cyst with the aim of treating the patient based on the final diagnosis.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

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