

Case Report

Second branchial cleft cyst in adolescent: a case report

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Received: 12 November 2025

Accepted: 27 November 2025

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ABSTRACT

Branchial cleft cysts are congenital lateral neck masses which manifest in the adolescents or in adulthood. They arise from the remnants of the branchial apparatus of embryonic life. Cystic remnants present commonly in the adolescence. Second branchial cleft cysts are the most common form of the branchial anomalies and originate from the remnants of the cervical sinus of His and its duct during the differentiation of branchial apparatus. Purpose of the study was to report a case of second branchial cleft cyst in adolescent. Patient was presented with second branchial cleft cyst at the age of 20 years. Computed tomography of neck showed well-circumscribed cystic lesion with an internal solid component in the peri vertebral space extending to the left submandibular space at the level of lower cervical region, attached to left sternocleidomastoid muscle causing minimal displacement of left internal and external carotid arteries medially and left internal jugular vein laterally, suggesting branchial cleft cyst. Complete surgical excision of the cyst had been done. The histopathologic study revealed a squamous epithelium-lined cyst with lymphoid infiltration, consistent with branchial cleft cyst. The patient was discharged without any complications. Clinical manifestations combined with knowledge of the embryology and spatial anatomy of the head and neck provide clues for a correct diagnosis and appropriate management of branchial cleft cyst.

Keywords: Branchial cleft cyst, Adolescent, Excision

INTRODUCTION

Congenital malformations (CM) are altered structures and functions of a newborn's organ systems that occur in intrauterine life and are identified before, at, or shortly after birth. The prevalence of CM varies from region to region, but the global prevalence of CM is approximately 2-3%.¹⁻³ One type of congenital malformation is the branchial cleft cyst.

Branchial cleft cyst is a congenital anomaly arising from the first to fourth pharyngeal pouches.^{4,5} These anomalies may present as fistulas, cysts, or sinus tracts, depending on the degree of incomplete obliteration during embryogenesis.^{6,7} The branchial arch represents the

embryologic precursor of the face, neck, and pharynx. Branchial arch anomalies are children's second most common congenital lesions of the head and neck. Branchial cleft cysts are CM arising from incomplete involution of the branchial remnant. About 90-95% commonly present at the age of 20 to 40 years.⁸

A branchial fissure cyst is believed to result from the failure of fusion of the second branchial arch. However, some theories suggest that cystic degeneration of the first, second, and third branchial fissures or cystic degeneration of epithelial elements of the neck lymph nodes may also lead to cyst formation. Branchial cleft cysts are more likely to affect women than men and usually present in the 3rd decade of life as a non-tender swelling on the

lateral side of the inferior neck of the mandible. They may increase in size, especially after upper respiratory tract infections, and sometimes show signs of inflammation and abscess formation such as pain, swelling, and edema. Acute enlargement may cause obstructive symptoms such as respiratory distress or dysphagia. Sometimes the cysts are associated with sinuses or fistulas related to the sternocleidomastoid muscle.^{9,10} In this case report we report a patient with a second branchial cleft cyst found at Prof. Dr. I. G. N. G. Ngoerah Central General Hospital Denpasar.

CASE REPORT

A 20-year-old female patient came to the ENT clinic of Prof. I. G. N. G. Ngoerah Hospital with a main complaint of a lump on the left neck 4 months before being hospitalized. The patient said the lump felt enlarged slowly. The lump does not feel painful. Previous complaints of fever did not exist, cough and cold were denied, and history of discharge from the ear was denied. Other complaints such as nasal congestion, double vision, nosebleeds, and headache are denied.

On physical examination, the impression was that the patient's general condition was compos mentis and nutritionally adequate. Based on physical examination in the left neck region, there was a soft and mobile palpable mass measuring 2×5 cm was found, there was no tenderness, as high as level IV, along the sternocleidomastoideus muscle.

Furthermore, a neck ultrasound supporting examination was performed with the results of a cystic lesion with a solid lobulated component and calcification in the left colli region posterolateral aspect suspected inflammatory process and cystic hygroma, multiple atypical lymphadenopathies right submandibular region left, right thyroid left and isthmus did not appear abnormal. Other supporting examinations performed were FNAB colli sinistra with cytomorphologic results of a benign cystic lesion impression.

CT scan examination of colli with contrast showed a cystic lesion with a solid component inside, well-defined, lobulated edges, septa in the perivertebral space, left submandibular space as high as the corpus vertebrae C2-6, measuring $\pm 1.5 \times 4.4 \times 6.7$ cm, which on contrast administration appears slightly contrast enhancement on the solid component and septa, the lesion appears attached to the left sternocleidomastoideus muscle, minimally pressing the left internal and external carotid arteries to the medial and left internal jugular vein to the lateral. A contrast-enhanced CT scan of the colli suggested a benign cystic lesion, suspected of branchial cleft cyst. Laboratory tests showed the following: Hemoglobin level, leukocyte count, platelet count, liver function, and renal function were normal. Chest X-ray results were normal.

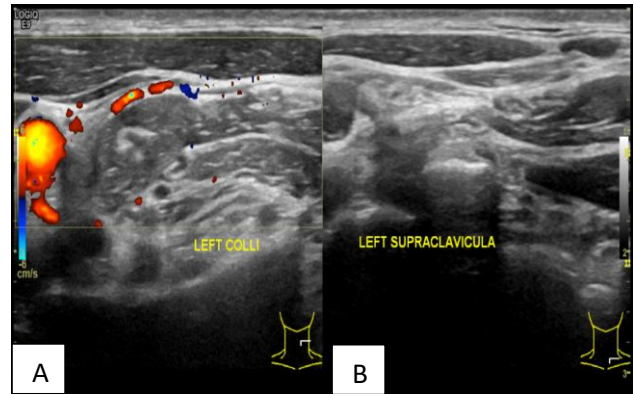


Figure 1 (A and B): Neck ultrasound of the patient.

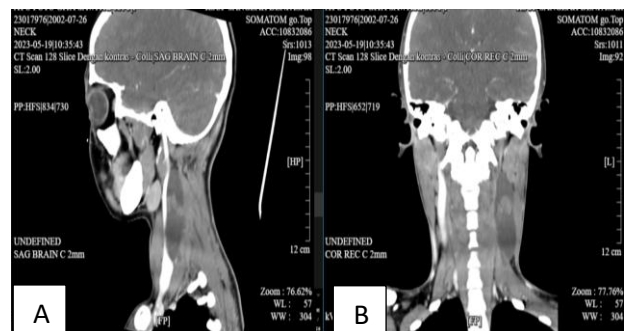


Figure 2 (A and B): CT scan of the patient.

The patient was diagnosed with a left branchial cleft cyst. The patient underwent left branchial cyst extirpation on July 6th, 2023, with operative findings of a cystic mass on the inferior sternocleidomastoideus muscle to the left internal carotid artery with a size of 4×4×10 cm. Histopathologic examination of the tissue showed tissue with a cystic structure lined with stratified squamous epithelium without signs of atypia. Some of the epithelium appeared attenuated. The cyst wall was a puffy connective tissue stroma containing lymphocyte aggregates. In other tissue sections, it is a lymphnode structure lined with fibrous connective tissue capsules, with diffuse proliferation of lymphoid cells, some of which appear atypic, with a fair number of lymphocytes among them. Histomorphology tended to be a branchial cleft cyst. On July 20th 2023, the patient came for wound care. The patient had no complaints and the wound was well-maintained.

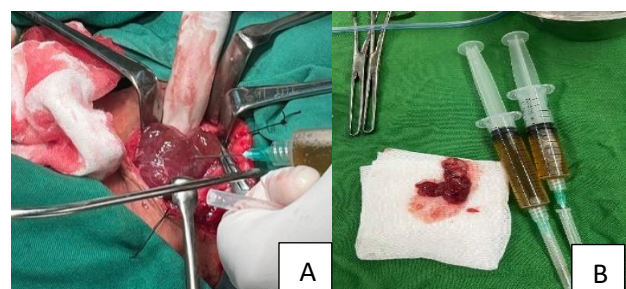


Figure 3 (A and B): Surgical findings.



Figure 4: The wound condition 2 weeks after surgery.

DISCUSSION

Branchial cleft cysts are congenital anomalies arising from the first to fourth pharyngeal clefts. The most common type of branchial cleft cyst arises from the second cleft, with anomalies originating from the first, third, and fourth clefts being less common.^{11,12} Branchial cleft cysts are a common cause of soft tissue swelling in the neck of young adults. It generally occurs unilaterally and is usually seen on the lateral aspect of the neck. It is clinically apparent in late childhood or early adulthood.⁷ The second branchial cleft anomaly most commonly presents as a cyst followed by a sinus and fistula. Most are present within the submandibular space but can occur anywhere along the course of the second branchial arch which extends from the skin over the supraclavicular fossa, between the internal and external carotid arteries, to enter the pharynx at the level of the tonsillar fossa. Fistulas or cysts in the lower anterior or lateral region of the neck most likely represent second branchial cleft anomalies.⁶

A fistula or cyst in the anterior or lower lateral region of the neck most likely represents an anomaly of the second branchial cleft. Fistulas are usually diagnosed in infancy/childhood by drainage of secretions or purulent material from an opening at the anterior border of the sternocleidomastoid in the lower third of the neck. The cyst is most commonly diagnosed as a painless lateral neck mass that is tender and/or increases in size especially if infected. Histologically, the cyst is filled with a cloudy yellow fluid containing cholesterol crystals and lined by stratified squamous epithelium. In adult patients, the main diagnostic consideration is if the cystic lesion is a metastatic lymph node and subsequent imaging is directed towards identifying the primary neoplastic lesion.^{6,8}

The history obtained from the patient is usually a complaint of a single mass in the neck, with or without an increase in size. Patients may experience compression

symptoms such as dyspnea, dysphagia, or even snoring if the mass extends to adjacent structures. Physical examination usually reveals a soft mass, with no tenderness and no fluctuation.⁹ Physical examination will differ depending on the location of the branchial cleft cyst. The second branchial cleft cyst is usually identified at the lower anterior border of the sternocleidomastoid and may connect to the tonsillar fossa of the pharynx. This type may be adjacent to the glossopharyngeal and hypoglossal nerves and carotid vessels. The cyst is tender if secondarily infected, and in severe cases, may cause airway compromise.¹⁶ On histopathologic examination, the lining of branchial cysts is generally squamous epithelium but sometimes pseudostratified, columnar, and ciliated. The lining may be ulcerated. The connective tissue wall contains an abundance of lymphoid tissue, suggesting germinal centers.⁷

The clinical findings in our case match the characteristics mentioned in the literature such as a lump on the left neck that is felt to be enlarging slowly. The lump is not painful. On physical examination of regio colli sinistra, a soft and mobile palpable mass measuring 2×5 cm, no tenderness, as high as level IV, along the sternocleidomastoideus muscle was found. These clinical findings are also supported by supporting examinations such as CT scans which show the results of a cystic lesion with a solid component in it, well-demarcated, lobulated edges, septa in the perivertebral space, left submandibular space as high as the corpus vertebrae C2-6, measuring ±1.5×4.4×6.7 cm, which on contrast administration showed slight contrast enhancement in the solid and septal components, the lesion was attached to the left sternocleidomastoideus muscle, minimally urging the left internal and external carotid artery medially and the left internal jugular vein laterally, suggesting a benign cystic lesion, suspected of the branchial cleft cyst.

The treatment of branchial cleft cysts is elective excision due to the very low risk of infection, further enlargement, or risk of malignancy. As long as there is no obvious airway obstruction or abscess, there is generally no urgency; thus, excision can be delayed until beyond 3 to 6 months of age. However, if there is airway obstruction or a large abscess, emergency surgery may be required.¹⁴ Excision is planned to optimize cosmetics, placing it within the natural skin fold if possible. If a fistula or sinus is present, then identifying the tract by carefully inserting a probe or catheter is important to ensure complete excision and reduce the chance of recurrence. Methylene blue can be used by dipping a lacrimal probe into the solution and inserting it into the duct to make it easier to identify intraoperatively. Dissection should be performed carefully on the lesion surface as the duct may be thin-walled. If the patient is unable to undergo surgery, ethanol ablation has been used as an alternative in this patient population, although it is not usually recommended as a primary treatment.¹⁸

Once a branchial cleft cyst is excised, recurrence is relatively rare. There is an estimated risk of 3%. Patients and families should be educated that branchial cleft cysts are usually benign, and with treatment, patients generally recover without complications or recurrence.^{7,16} In our case, significant improvement in symptoms was observed after excision. The patient's postoperative condition and wound were well maintained. No complications such as infection occurred.

CONCLUSION

Branchial cleft cysts are CM, and currently, there are no preventive measures to reduce the chance of occurrence. Patients and healthcare professionals should be educated about the symptoms and physical examination findings that can lead to an early diagnosis of these lesions so that they can be treated quickly and appropriately. Proper management of branchial cleft cysts can improve the patient's quality of life, with low recurrence rates and very rare complications.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Ketut SI, Made JI, Gde ANI, Wayan LLI, Ayu AWI. Second branchial cleft cyst in adolescent: a case report. *Int J Otorhinolaryngol Head Neck Surg* 2026;12:79-82.