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Branchial Cysts: A Case Series of Surgical Excision of These Embryological Neck Enigmas.

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Abstract

Branchial cleft cysts also known as branchial cleft anomalies are congenital anomalies arising from the pharyngeal clefts. The true incidence of these anomalies is unknown despite their relative frequency. which is likely due to the variety in the anomaly's presentation as well as the occurrence of complications that cause errors in accurate reporting. We present a retrospective case series of seven cases of patients with branchial cysts referred to and operated upon at a tertiary care centre for Otorhinolaryngology in India from 2021-2022. The case series was formulated of patients diagnosed with branchial cysts (through history, clinical examination, Ultrasonography, Contrast Enhanced Computed Tomography and Fine Needle Aspiration Cytology) and referred to the Department of Otorhinolaryngology for management. The clinical presentation in all cases was a lateral neck mass of insidious onset and gradual progression not associated with pain or obstructive symptoms. Histopathologically, all cysts showed different constituents. All patients underwent surgical excision and no patient had recurrence or complications. The clinical evaluation of such cases followed by radiological confirmation with USG/CT/MRI and FNAC for histopathological confirmation is the mainstay for the diagnosis of such cysts. Treatment remains surgical excision and care must be towards preventing damage to surrounding neurovascular/muscular structures.

Keywords: branchial cyst, branchial cleft anomaly, congenital neck mass, lateral neck mass, excision of branchial cysts, branchial cleft cysts

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INTRODUCTION

The term 'branchial' originated in the Greek language from the word 'bragchia' meaning 'gills' [1]. Pharyngeal arches develop at week 4 of embryonic development just lateral to the stomodeum and above the cardiac bulge. These well-defined outgrowths are six in number (the fifth arch regresses spontaneously) and are a proliferation of mesoderm germ layer infiltrated by neural crest cells.

These arches result in the emergence and development of numerous head, neck and facial structures over the course of embryonic development. Malformations include cysts, fistulas and sinuses which occur as a consequence of incomplete obliteration of the clefts and pouches that separate the arches. The second arch is the most common site for these anomalies (>95 % of all branchial anomaly cases) [2].

There are many theories and explanations given for the formation of branchial cysts, however, none have been proven to be conclusive to date [3,4]. These include:

- Cystic degeneration of branchial clefts.
- Failure of fusion at branchial arches (especially the second arch).
- Cystic degeneration of cervical lymph nodes (epithelial elements).

These cysts are more common in the female population, especially between 20-30 years of age [3]. They present as a non-tender swelling in the neck (lateral aspect). Acute enlargement can cause dysphagia or respiratory dysfunction. Pain also may be associated in these cases in the event of abscess formation or inflammation. Diagnosis can be confirmed with imaging modalities like Ultrasonography (USG)/Computed Tomography (CT)/Magnetic Resonance Imaging (MRI) and further with Fine Needle Aspiration Cytology (FNAC). Treatment is mainly in the form of excision of cysts [4].

MATERIALS AND METHODS

A retrospective case series of patients diagnosed with branchial cysts (through history, clinical examination, Ultrasonography, Contrast-Enhanced Computed Tomography (CECT) and Fine Needle Aspiration Cytology (FNAC) and referred to the Department of Otorhinolaryngology for management was undertaken.

RESULTS

Case Report 1

32-year-old male reported a right-sided upper neck swelling of 03 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 5 cm × 5 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the submandibular region of the right side (superior extent – tip of mastoid, inferior extent - hyoid, anterior extent – the angle of the mandible, posterior extent - deep to the right Sternocleidomastoid muscle (SCM)). The skin over the swelling was normal with no local rise in temperature. Ultrasonography revealed a 2.7 cm × 4.2 cm × 4.3 cm well-defined cystic lesion in the submandibular region (right side) lying on the right common carotid artery (lateral aspect) and extending cephalad from the bifurcation. CT findings were of a 4.6 cm × 4.0 cm × 3.2 cm cystic swelling in the right submandibular region.

USG-guided FNAC revealed no malignant/atypical cells with scattered neutrophils and proteinaceous substances. The patient was given a provisional diagnosis of a right branchial cyst.

The cyst was excised under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below right-side mandible (Figure 1)
- Superior and inferior skin and sub-platysmal flaps raised (Figure 2)
- Spinal accessory nerve passing over cyst separated carefully by dissection (Figure 3)
- Cyst removed in totality
- Wound closed in layers

The postoperative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

There was no recurrence or other complications on six months follow-up.

Figure 1: Case Report 1 - Skin incision

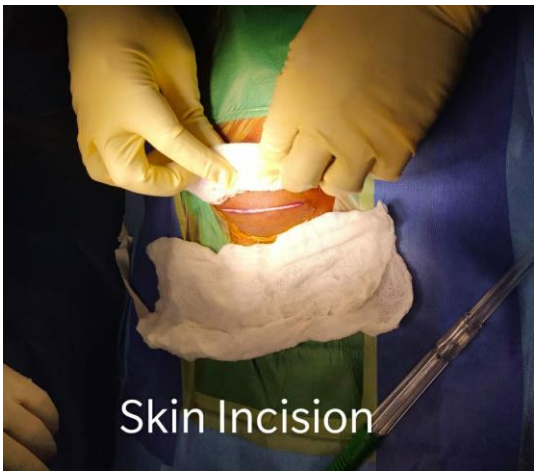


Figure 2: Case Report 1 - Subplatysmal flap elevation

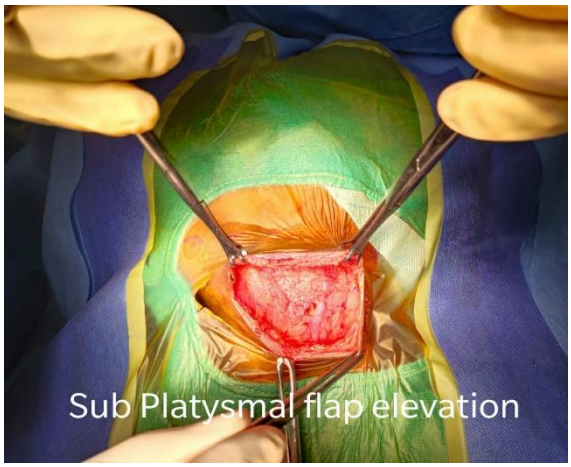
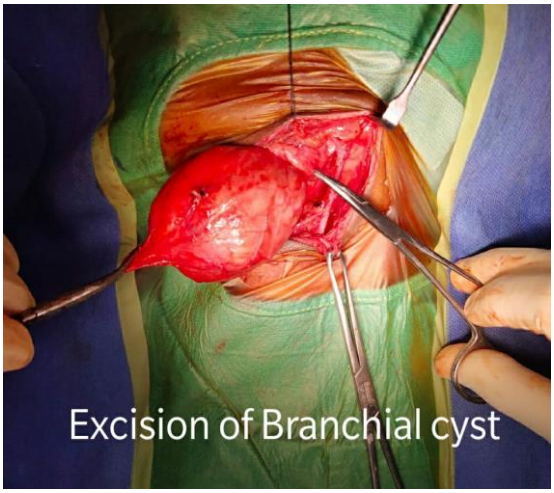


Figure 3: Case Report 1 - Excision of branchial cyst



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Case Report 2

A 14-year-old male presented with a left-sided upper neck swelling of 02 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 4 cm × 2 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the left anterior triangle. The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 3.2 cm × 4.6 cm × 4.9 cm well-defined lesion with cystic characteristics in the left anterior triangle.

findings were of a 3.5 cm × 4.6 cm × 5.2 cm cystic swelling in the left anterior triangle region.

USG-guided FNAC showed proteinaceous smears with numerous foamy macrophages, many containing hemosiderin. The patient was given a provisional diagnosis of a left branchial cyst.

The branchial cyst was excised under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below left side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Cyst removed in totality by separating it from surrounding fascial planes (Figure 4, 5)
- Wound closed in layers

The postoperative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

There was no recurrence or other complications on six months follow-up.

Figure 4: Case report 2 - Cyst visualized intraoperatively

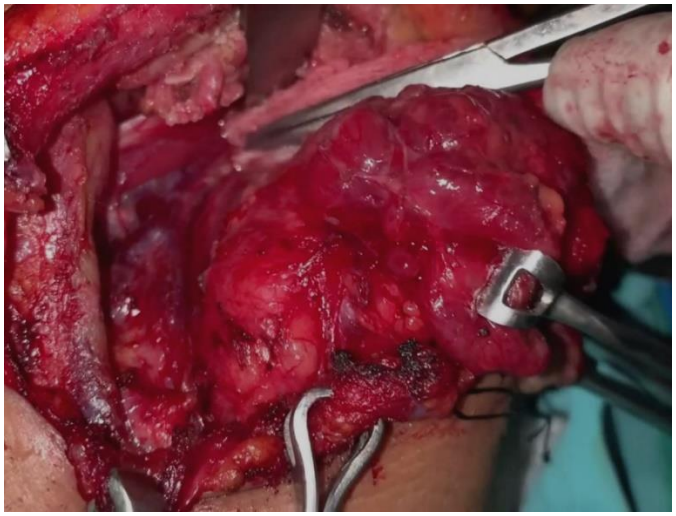

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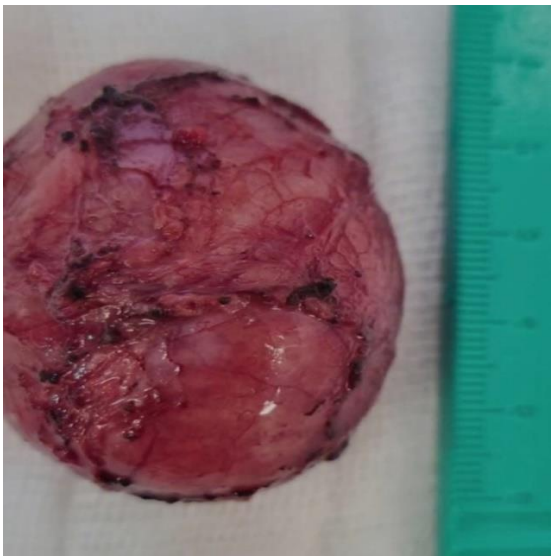
Figure 5: Case Report 2 - Specimen



Case Report 3

A 25-year-old female presented with a right-sided upper neck swelling of 02 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 5 cm × 6 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the right anterior triangle. The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 5.2 cm × 4.6 cm × 6.2 cm well-defined lesion (cystic character) in the right anterior triangle.

CT findings were of a 5.1 cm × 4.4 cm × 6.2 cm cystic swelling in the right anterior triangle region.

USG-guided FNAC revealed abundant macrophages and lymphocytes, squamous epithelial cells of varied maturity, on a backdrop of amorphous debris. The provisional diagnosis given was of a right branchial cyst.

The cyst was removed under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below right side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Cyst dissected carefully from the fascia of anterior and inner aspects of SCM (Figure 6)
- Cyst removed in totality (Figure 7)
- Wound closed in layers

The post-operative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

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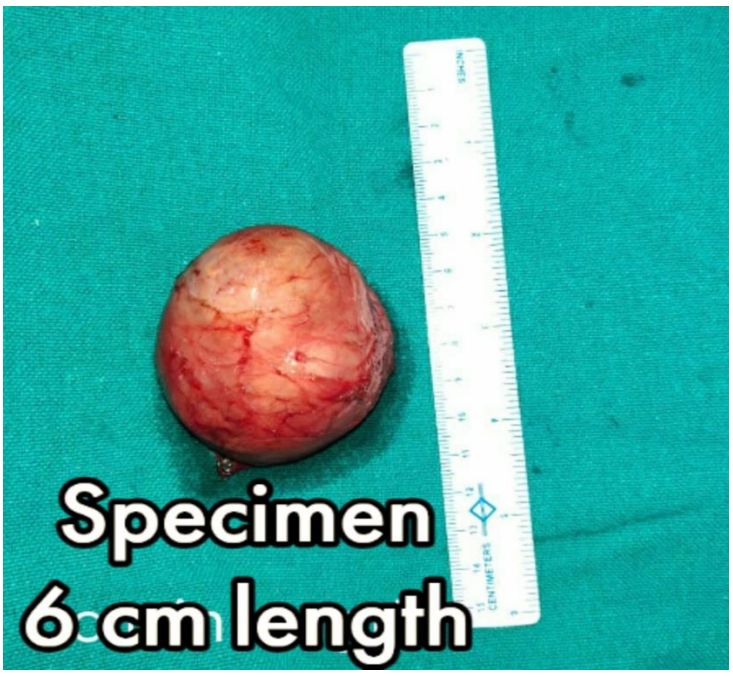
Figure 6: Case Report 3 - Cyst seen intraoperatively

Figure 7: Case Report 3 - Specimen



Case Report 4

An 8-year-old male presented with his parents in Out Patient Department with a right-sided upper neck swelling of 02 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 3 cm × 3 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the submandibular region (right side) (superior extent – the mastoid tip, inferior extent - hyoid, anterior extent - the angle of the mandible, posterior extent - deep to the right SCM). The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 2.7 cm × 2.2 cm × 3.3 cm well-defined cystic lesion in the submandibular region (of the right side) lying on the lateral aspect of the right common carotid artery and extending cephalad from the bifurcation. CT findings were of a 2.6 cm × 2.3 cm × 3.2 cm cystic swelling in the right submandibular region.

USG-guided FNAC revealed scattered inflammatory cells of mixed populations which included numerous lymphocytes and foamy macrophages. The patient was given a provisional diagnosis of a right branchial cyst.

The cyst was removed under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below right side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Cyst dissected carefully from surrounding soft tissue structures (Figure 8)
- Cyst removed in totality (Figure 9)
- Wound closed in layers

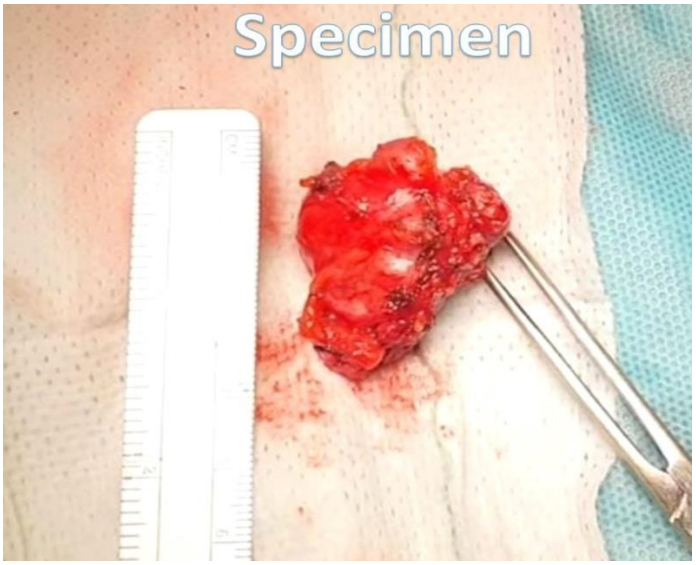
The post-operative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

There was no recurrence or other complications on six months follow-up.

Figure 8: Case Report 4 - Cyst seen intraoperatively

Figure 9: Case report 4 - Specimen



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Case Report 5

A 10-year-old male reported with a right-sided upper neck swelling of 03 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 3 cm × 3 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the submandibular region (right) (extending superiorly from the mastoid tip and right up till the hyoid inferiorly, anterior from the mandible (angle of mandible) to posteriorly deep to the right SCM). The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 2.7 cm × 3.1 cm × 3.3 cm well-defined cystic lesion in the submandibular region (right side) lying on the right common carotid artery (its lateral aspect) and extending cephalad from the bifurcation. CT findings were of a 2.6 cm × 3.2 cm × 3.4 cm cystic swelling in the submandibular region (right).

USG-guided FNAC revealed mature superficial squamous cells with intact nuclei, anucleate clusters of neutrophils and lymphocytes, macrophages and amorphous debris in the background. The patient was given a provisional diagnosis of a right branchial cyst.

The cyst was excised under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below right side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Cyst separated carefully by dissection from surrounding structures (Figure 10)
- Cyst removed in totality (Figure 11)
- Wound closed in layers

The postoperative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

There was no recurrence or other complications on six months follow-up.

Figure 10: Case report 5 - Cyst seen intraoperatively

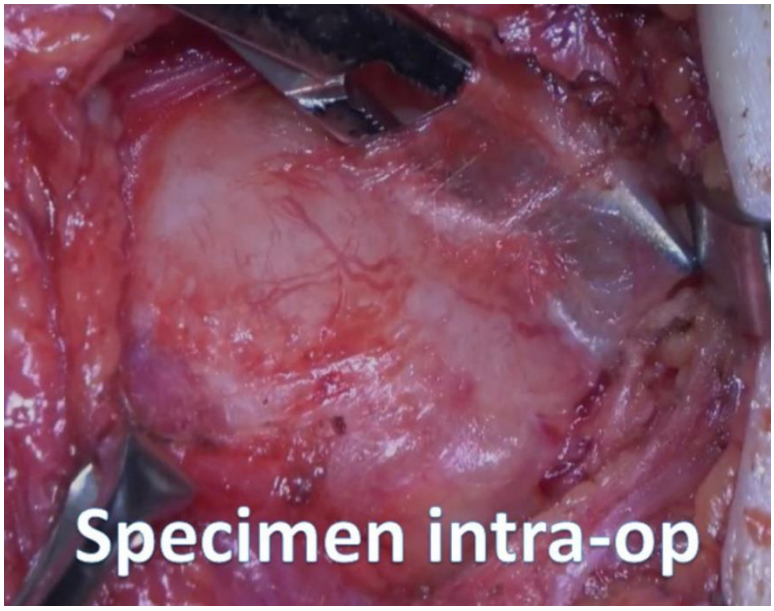

Figure 11: Case report 5 - Specimen

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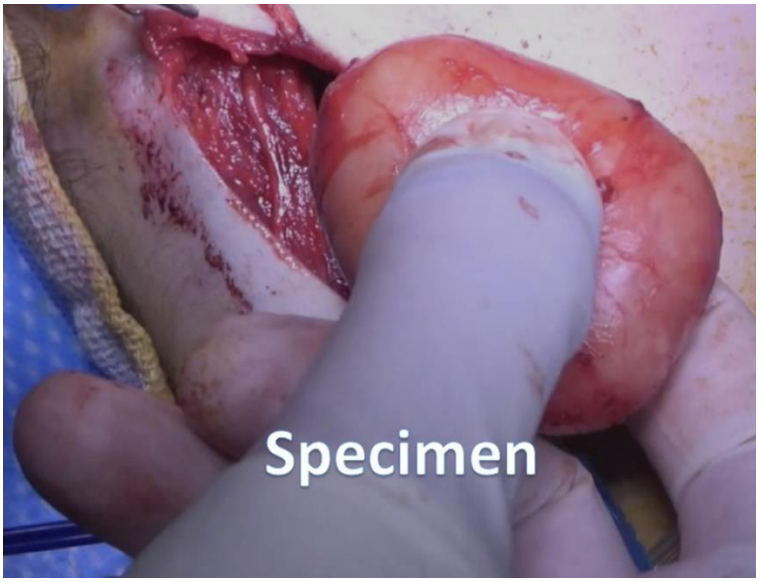

Case Report 6

A 17-year-old female presented with a left-sided upper neck swelling of 02 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 7 cm × 8 cm globular, soft, non-tender, cystic, compre0ssible, non-transilluminant, mobile swelling extending in the left anterior triangle. The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 7.2 cm × 6.6 cm × 7.3 cm well-defined lesion (cystic) in the left anterior triangle.

CT findings were of a 7.1 cm × 6.5 cm × 7.4 cm cystic swelling in the left anterior triangle region.

USG-guided FNAC showed anucleated and nucleated squamous cells with scattered inflammatory cells on a dirty background. The provisional diagnosis was that of a left branchial cyst.

The cyst was removed under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below left side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Surrounding structures separated carefully by dissection away from the cyst (Figure 12,13)
- Cyst removed in totality (Figure 14)
- Wound closed in layers

The postoperative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

was no recurrence or other complications on six months follow-up.

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Figure 12: Case Report 6 - Cyst seen intraoperatively

Figure 13: Case Report 6 - Intraoperative picture after cyst dissection

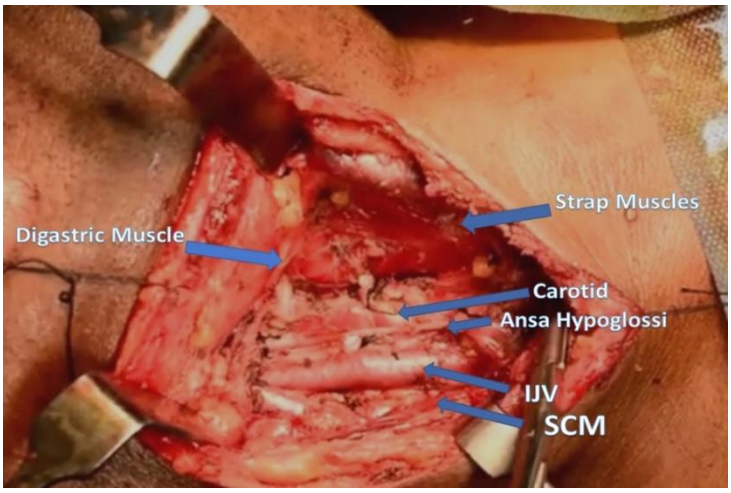
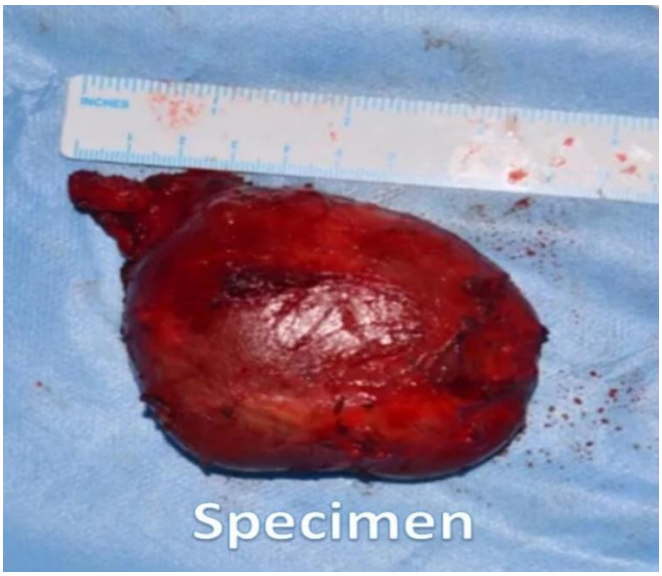


Figure 14: Case Report 6 - Specimen



Case Report 7

A 9-year-old male reported with guardians to outpatient department with a right-sided upper neck swelling of 05 months duration which was painless, progressive and not associated with pressure symptoms (voice change/difficulty in deglutition or breathing). Examination revealed a 2 cm × 3 cm globular, soft, non-tender, cystic, compressible, non-transilluminant, mobile swelling extending in the submandibular region (right) (superior extent - mastoid tip, inferior extent – hyoid, anterior from the angle of the mandible to posteriorly deep to the right SCM). The skin over the swelling was normal with no local rise in temperature.

Ultrasonography revealed a 5.3 cm × 3.2 cm × 5.3 cm well-defined cystic lesion in the submandibular region (right) lying on the right common carotid artery (its lateral aspect) and extending cephalad from the bifurcation. CT findings were of a 5.2 cm × 3.3 cm × 5.2 cm cystic swelling in the submandibular region (right side).

USG-guided FNAC revealed sheets of lymphocytes, squamous cells and respiratory epithelium. The Provisional diagnosis was of a right branchial cyst.
The cyst was surgically removed under GA (General Anaesthesia). The steps were as follows:

- Skin incision 2 cm below right side mandible
- Superior and inferior skin and sub-platysmal flaps raised
- Spinal accessory nerve passing over cyst separated carefully by dissection (Figure 15)
- Cyst removed in totality (Figure 16)
- Wound closed in layers

The postoperative period of the patient was uneventful.

Histological examination revealed the swelling to be a branchial cyst.

There was no recurrence or other complications on six months follow-up.

Figure 15: Case report 7 - Cyst seen intraoperatively

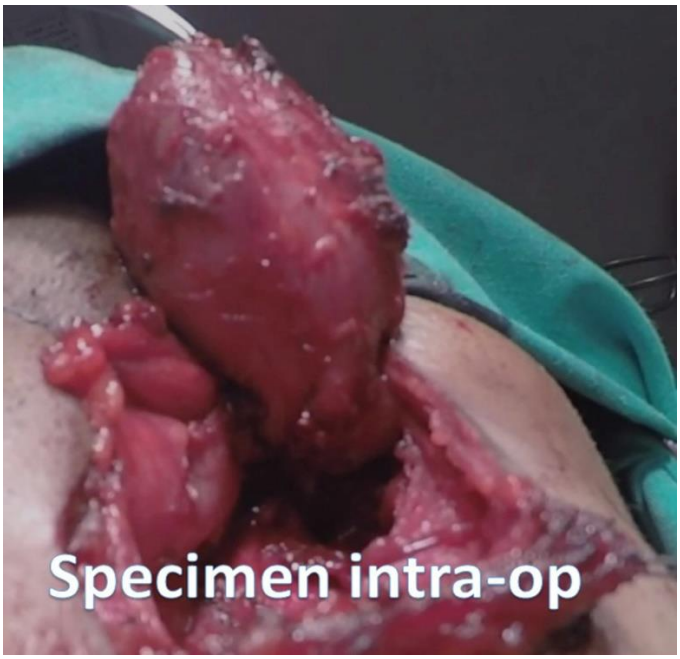


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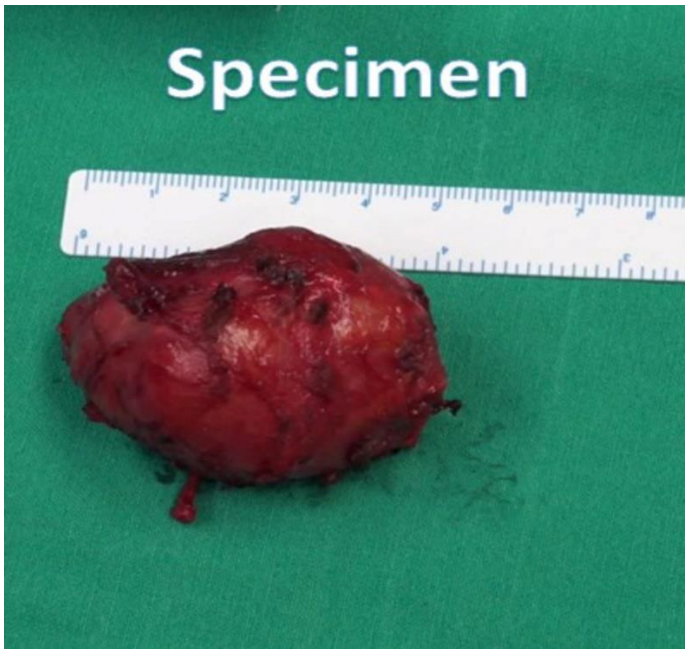
Figure 16: Case report 7 - Specimen



DISCUSSION

In the 18th century, the Austrian surgeon Hunczovsky, had given the first detailed record of the occurrence of lateral cysts of the neck [5]. 'Branchial cyst' as a term was first utilized by Ascherson in 1832 when he described his 'Branchial Theory' [6]. Various theories explain its origin [1].

- Branchial apparatus theory - Branchial cysts are formed as remnants of the pharyngeal pouches or the branchial clefts (or as a result of their fusion). This however doesn't explain the incidence in young adults instead of at birth.
- Cervical sinus theory – Branchial cysts develop from the remnants of the cervical sinus of His. This sinus is formed when the second arch grows down and fuses with the 5th arch.
- Thymopharyngeal duct theory - These cysts are remnants of the connection between the third branchial pouch and the thymus.
- Inclusion theory - These are inclusions (epithelial variety of inclusions) within a lymph node.

Types of Branchial Cysts

First Branchial Cleft Cyst

First cleft cysts comprise approximately 5% to 25% of all branchial cleft anomalies. These anomalies are further subclassified via the Work classification system.

Work type I: These cysts contain only ectoderm. On clinical examination, they show pre-auricular masses/sinuses that track anteromedially to the external auditory canal. They frequently present laterally to the facial nerve and end within the confines of the external auditory canal. They may also connect to the middle ear (umbo), essentially occurring as an external auditory canal duplication.

Work type II: The kinds of cysts are more common. They contain both mesoderm and ectoderm. These classically present within the submandibular triangle (or at the angle of mandible). Their course may be medial/lateral to the facial nerve and pass deep (in 30% cases), superficial (in 57% cases) or in between (13% cases) branches of the facial nerve [7].

Second Branchial Cleft Cyst

These are the most commonly occurring branchial cleft cysts. They comprise an estimated 40-95% of branchial anomalies. The cyst's external punctum is found anteromedial to the SCM on the neck's skin. Branchio-oto-renal syndrome can also have the occurrence of second branchial cleft cysts that occur bilaterally. The cyst's external opening is located on the neck's skin. The cyst's fistula is located deep to the platysma and then passes in between the carotids (internal and external carotid arteries). It further courses superficial to both the hypoglossal as well as glossopharyngeal nerves and finally connects with the tonsillar fossa. Cysts or sinuses can develop anywhere along this course [8, 9].

Third Branchial Cleft Cyst

These cysts comprise an estimated 2-8% of all branchial cleft anomalies. In these cases, the external opening is seen over the skin over the anterior SCM (the middle third to lower third part). The course of the sinus follows starting at the skin opening as described above. Further beyond the skin opening, the tract then courses from deep to the platysma and later posterior to the carotid (internal carotid artery). The tract passes between the hypoglossal and glossopharyngeal nerves. It may also be closely associated with the superior laryngeal nerve and commonly it is seen that the tract courses superior to the nerve. It finally reaches the larynx (pyriform sinus) [10].

Fourth Branchial Cleft Cyst

These kinds of cysts are very rare. They comprise only 1% of overall branchial cleft anomalies. They occur more on the left side. The SCM (its medial lower border) hosts the skin opening for the anomaly. The exact course isn't well described due to its rarity. However, it is classically described to pass deep to the common carotid and further along its course it can loop around either the subclavian (in a right-sided anomaly) or the aortic arch (in a left-sided anomaly). These anomalies course superficial to the hypoglossal nerve and recurrent laryngeal nerve, and terminate the course in the larynx (at the apex of the pyriform sinus) [1].

Second branchial cleft cysts are also known as lateral cervical cysts. They lie deep to the sternocleidomastoid muscle (anterior border). They are also most commonly found at the junction of the upper and lower third of the SCM. They can however develop anywhere along the course of the second branchial cleft fistula, from the skin on the lateral aspect of the neck to the palatine tonsils, going between the carotid arteries (external and internal carotid arteries) [5]. They are usually asymptomatic and on clinical examination are found to be soft, non-tender, round, smooth, mobile, and fluctuant masses. They are also mostly covered with normal skin (unless they are complicated with infection, etc.). They are also usually slow growing and take many weeks (even years) to attain full size at clinical presentation. Infected cysts can also present with pain and may rupture due to the formation of an abscess leading to the formation of fistulae later. The cyst wall contains lymphoid tissue which enlarges during upper respiratory tract infections resulting in secondary enlargement of the cyst. Depending on size and location, they can cause cosmetic deformity, dysphonia, dysphagia and dyspnea [1, 11].

Carcinomas of Branchial cleft cysts are extremely rare entities [5].

Branchial cysts are diagnosed with history, clinical examination, Ultrasonography, CT/MRI and USG-guided FNAC [12]. CECT and MRI are superior to USG in the diagnosis of the cyst while also providing information on the organ of origin, nature and relation with neighbouring structures, volume and other characteristics thus helping better plan surgery and minimize surgical complications. Ultrasonography is a however cheaper, non-invasive modality but may fail to distinguish it from other cystic neck lesions [13].

Differential diagnoses for these conditions include [14]

- Lymphadenopathy
- Hemangioma
- Carotid Body Tumour
- Cystic Hygroma
- Ectopic thyroid/salivary tissue
- Vascular malformation/neoplasm
- Thyroglossal duct cysts
- Cat scratch disease

- Atypical mycobacterial infections
- Cystic squamous cell carcinoma

The treatment of choice is complete surgical resection of the cyst while preserving surrounding structures.

Important vascular structures like the carotid arteries (internal and external) and nerves like the vagus, hypoglossal, glossopharyngeal and superior laryngeal nerves must be identified to avoid injury.

Surgical complications include recurrence. The estimated risk is 3% of all cases. In cases in which there has been previous surgery or recurrent infection, recurrence can occur in up to 20% of cases [15].

Other complications include persistent fistula, and damage to the cranial nerves [5]. In one of our cases, the spinal accessory nerve was passing just over the branchial cyst and this warranted a cautious dissection to prevent nerve injury.

CONCLUSIONS

In India and worldwide, branchial cysts cases are very uncommon, and the true incidence of the same remains unknown. The clinical evaluation followed by radiological confirmation with USG/CT/MRI and FNAC for histopathological confirmation is the mainstay for the diagnosis of such cysts. Treatment remains surgical excision and care must be towards preventing damage to surrounding neurovascular and muscular structures while removing the cyst in totality to prevent recurrence.

Conflict of Interest: No conflict of interest noted. No financial implication.

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