

Case Report

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A Rare Presentation of Second Branchial Cleft Cysts in Adult Patient: Case Report with Literature Review

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ABSTRACT

Second branchial cleft cysts are the most common type of branchial cleft cysts as they make up approximately 95% of all cases. These are congenital conditions that occur in the upper lateral neck just anterior to the sternocleidomastoid muscle and caused by fluids filling spaces during early embryonic development. We present a case of atypical presentation of second branchial cleft cysts in 40 year's patient. A surgical intervention was performed with complete excision of the cysts.

Many of branchial cleft cysts would go unnoticed. However, upper respiratory tract infection can cause a sudden and significant increase in the size of the cyst because of the lymphoid tissue beneath the epithelium. The Differential diagnosis would include lymphadenopathy, lipoma, nerve sheath tumor, inflammatory lesions, carotid body tumor, external laryngocele, cystic hygroma, metastatic squamous cell carcinoma, tuberculosis-related, HIV-related lymphadenopathy, sarcoidosis, cat-scratch disease, lymphoma, and papillary thyroid carcinoma metastasis.

Surgical excision is the only known treatment for second branchial cleft cysts; moreover, a complete removal of second branchial cyst cleft is of paramount importance since it is key to preventing any recurrence of the disease.

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Introduction

Second branchial cleft cysts are the most common type of branchial cleft cysts as they make up approximately 95% of all cases [1-4]. These are congenital conditions that occur in the upper lateral neck just anterior to the sternocleidomastoid muscle and caused by fluids filling spaces during early embryonic development.

The condition's aetiology remains unknown, although it is believed that they derived from branchial apparatus development and more specifically from the second branchial arch [5].

They are most often reported in adults, whereas sinuses, fistulas and cartilaginous remnants are more common in children [4].

Branchial cleft cyst, also called benign cervical lymphoepithelial cyst, has a controversial and disputed pathogenesis [5]. But recurrent inflammation near the mandibular angle is often synonymous of a second branchial cyst [3].

The cyst manifests itself as a slow-growing, fluctuant and swollen soft tissue in the lateral aspect of the neck. It may be bilateral in which case familial occurrence is often recorded [5].

Case report 1

A 40-year-old man presented to our hospital with complaints of a right-sided neck swelling during the last 8 months, no limitation on mouth opening, and no pain aggravated by palpation of the region. The patient also stated that he suffered mandibular trauma five years ago.

He reported that he had a history of unilateral "cysts" in the lower one-third of his neck and that he underwent cervicotomy a year earlier. He stated that this area feels like a palpable, swollen knot which is not painful to the touch. Three weeks after his first surgery the cyst came back.

On exam, the patient's vital signs were within normal limits. A physical examination did not reveal nerve paralysis, or hearing, facial, or neck sensation disturbances, but a mobile, not tender but compressible mass was detected in the right neck region at the anterior border of the sternocleidomastoid muscle, between the mandibular angle and the clavicle with normal skin overlying the swelling (Figure 1) without drainage. There was no dysphagia, dysphonia, or dyspnoea.



Figure 1: Right lateral neck mass

Ultrasound, which is the first-line imaging method of choice for defining the nature of a benign cystic lesion, revealed a mass of about $90 \times 31 \times 30$ mm, a sharply demarcated lesion with posterior acoustic enhancement associated with visible walls; the cyst was echoic with internal debris.

On CT scan there was a right latero cervical masse extended the sub angulomandibular angle to the supraclavicular hollow, with thin wall, not enhanced after injection of the contrast product, well limited, measuring 55.8×42 mm (Figure 2).

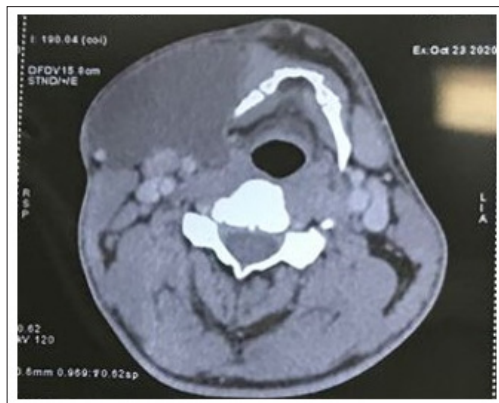


Figure 2: axial view of CT-scan showing second branchial cyst



Figure 3: longitudinal view of CT-scan showing second branchial cyst

A surgical intervention was performed under general anesthesia using a right transverse cervical approach. Incision was followed by exposure of the platysma with careful dissection of the surrounding structures (Figure 3)

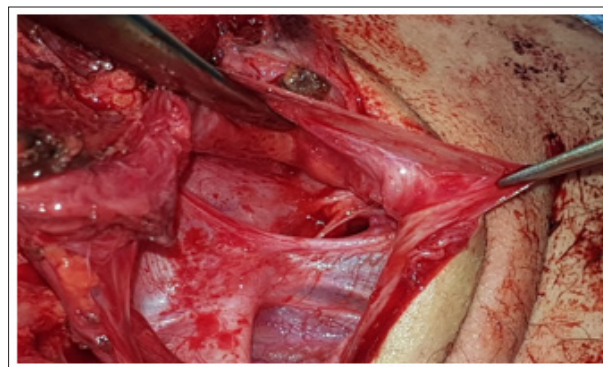


Figure 4: Intraoperative findings: superficial cyst, anterior to the sternocleidomastoid muscle (SCM)

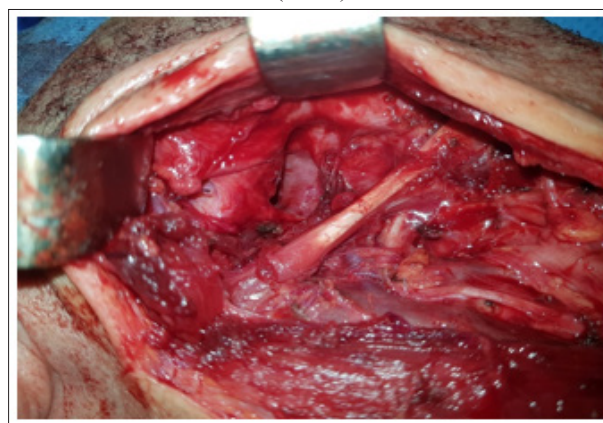


Figure 5: The lesion was removed completely

Discussion

Located at the anterior border of the sternocleidomastoid muscle between the mandibular angle and clavicle, second branchial cleft cysts are painless compressible swellings whose size can fluctuate over time. Most of the time these anomalies are unilateral, but when bilateral they are often combined with other congenital abnormalities. Branchial cleft cysts are benign and usually painless unless they are infected.

BCCs have no gender predilection [2]. However, a few studies reported that they occur more commonly in females [4]. While other authors are claiming that 60% of cases occur in males [6]. Many BCCs would go unnoticed. However, upper respiratory tract infection can cause a sudden and significant increase in the size of the cyst because of the lymphoid tissue beneath the epithelium [5]. Bilateral second branchial cleft cysts are very rare and unusual, but when present they can cause hearing impairment, cup-shaped ears, preauricular pits.

They are classified into four subtypes, by Bailey as follows [3, 6]:

- Type 1: situated anterior to the sternocleidomastoid, just deep to the platysma,
- Type 2: the most common variant of the four subtypes, found deep to the sternocleidomastoid, lateral to the carotid space,
- Type 3: these extend medially between the bifurcation of internal and external carotid arteries up to the lateral pharyngeal wall,
- Type 4: positioned in the pharyngeal mucosal space, medial to the carotid sheath

In most cases, a thorough physical exam is all that's needed to diagnose the condition when the cyst is in its usual and classic position [3]. But a Computed tomography (CT) scan, ultrasound, or MRI might be required to confirm the clinical diagnosis. On

USG, it looks like an anechoic mass or chiefly hypoechoic cystic mass, with faint internal debris and posterior enhancement. It looks like a pseudo-solid heterogeneous mass with internal debris and septa [3].

While preoperative ultrasound examination of the neck has many advantages such as simplicity, convenience, and rapidity, it fails to show the details of the anatomical structure around the cyst. On the other hand, Doppler ultrasonography with its 91.67% sensitivity, 50% specificity, and 90% accuracy has the potential to accurately diagnose and localize second BCC [7].

The Differential diagnosis would include lymphadenopathy, lipoma, nerve sheath tumor, inflammatory lesions, carotid body tumor, external laryngocele, cystic hygroma, metastatic squamous cell carcinoma, tuberculosis-related, HIV-related lymphadenopathy, sarcoidosis, cat-scratch disease, lymphoma, and papillary thyroid carcinoma metastasis [3, 8].

On a typical CT scan, the cyst usually appears as a well-circumscribed, non-enhancing mass of homogeneous low attenuation, with clear boundaries and a uniform surrounding thin and smooth wall [2, 3, 7]. Unless there's infection hemorrhaging or carcinomatous transformation, which makes diagnosis difficult. These images can be mildly enhanced by injecting contrast materials [9].

On MRI findings present that cystic fluid show low to slightly high signal intensity on T1WI and high signal intensity on T2W [7]. For a definitive diagnosis we need to proceed with surgical resection and biopsy since the second BCC has no specific clinical or imaging features.

The probability of a second branchial cleft cyst turning malignant is very low. Malignant transformation must be considered when BCCs exhibit ill-defined borders or thickened walls with adjacent infiltrating vessels, or cause lymphadenopathy atelectasis. The malignant transformation of BCCs is usually associated with recurrent infection, repeated surgery or biopsy and other carcinogenic stimuli. It is also highly important to consider and analyze the patient's history. Only a pathological examination after a surgical resection allows for a reliable and definitive identification of malignancies. Diagnoses based on clinical symptoms, imaging techniques or even biopsy often turn out difficult and flawed [9].

Surgical excision is the only known treatment for second branchial cleft cysts. Surgery should be performed only after all infections have been treated with antibiotics.

The traditional surgical approach which consists in a large cervical incision works just fine and ensures a complete removal of the cyst. But it has a major downfall since there is a large amount of intraoperative blood loss and results in a relatively significant and prominent scar. Alternatives to the open surgical method have been developed. Minimally invasive surgery techniques such as endoscopic surgery and retro auricular hairline incision, by avoiding cutting off the muscles, allow for quicker recovery time and less surgical trauma, less pain and discomfort and more cosmetic benefits [4,7].

To prevent any risk recurrence the surgeon must make sure the excision is complete. An ipsilateral tonsillectomy ensures removal of the fistulous tract [10]. But it is not required, as some studies have shown, to avoid recurrences [11].

Conclusion

A complete removal of second branchial cyst cleft is of paramount importance since it is key to preventing any recurrence of the disease. And this can be achieved more easily if the cyst contents are partially aspirated. It is also highly recommended to treat any infection of the cyst before surgery.

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