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Research Article

Unusual Presentation of a Large Multilocular Second Branchial Cleft Cyst

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Abstract

Second branchial cleft cysts are the most common type of branchial abnormalities and usually found high in the neck. Oropharyngeal presence of a branchial cleft cyst is very rare. We report a rare case of, oropharyngeal, second branchial, multilocular, cleft cyst in a nine-year-old child (8.0 x 5.0 cm in maximum diameter). The cyst was removed completely via extra / intra-oral approach and did not have tract-like structure. The anatomic location together with the histopathology results, which showed a squamous epithelium-lined cystic wall with lymphoid aggregation, were characteristic findings of a Branchial Cleft Cyst (BCC). Patient was discharged without any complication and a regular follow up, of 6 months, showed no evidence of re currency. Having reviewed the literature, it seems to be that this is the first case to be reported of a multilocular branchial cleft cyst, excised of this size, from the oropharyngeal area in a child. BCC shares a clinical presentation with other pathological entities of the neck, making diagnosis difficult at times. Health professionals should be aware of this entity because it can be easily confused with an odontogenic infection and submandibular or parotid gland pathology, especially if it develops high up in the neck.

Introduction

Branchial cleft anomalies may present as a cyst, sinus, fistula, or cartilaginous remnant [1]. Approximately 80% of branchial cleft anomalies present as a cyst [2] and about 95% are formed from the region of the second branchial arch [3,4]. The remaining 5% arise from the regions of the first, third, or fourth arches [3,4]. A second Branchial Cleft Cyst (BCC) typically presents as a painless, mobile, and fluctuant mass located along the anterior border of The Sternocleidomastoid Muscle (SCM), usually just above the clavicle [2]. Approximately 97% to 98% of the lesions are unilateral [5], and of these, 83% to 97% are on the left side, presumably, subsequent to asymmetrical vascular development [6]. Although branchial cleft cysts are congenital and might be noted at birth, most are not detected until the first or second decade of life [7]. Some are detected when they become more prominent in late childhood. Other cases become apparent when encountering upper respiratory tract infections or when the cyst becomes infected [8]. Although BCCs are congenital and might be noted at birth, most are not detected until the first or second decade of life [7]. Some are detected when they become more prominent in late childhood.

Case Report

A nine-year-old female child presented with a swelling located just below the jawline in the, lateral, left submandibular region of the neck. The swelling has been present since birth. The patient was referred to remove a mucocele, which was rapidly increasing in size over the last 2 months, prior to examination. There were no associated complaints such as pain, change in voice, or difficulty in breathing. The swelling was not moving with protrusion of tongue or on deglutition. Intra-orally, the swelling was located in the floor of the mouth along the left lateral border of the tongue and extends posteriorly to the oro-pharyngeal area, measuring approximately 1.0 cm × 3.0 cm. There was no history of discharge during eating or drinking. However, the patient gave a history of some fluid discharge from the lesion, occasionally, which could be the mucin present in the cyst or could be due to the infection. On examination, extra-orally, smooth, well-defined, non-tender cystic mass measuring approximately 7.0 cm × 5.0 cm was noted just below the jawline in the left lateral part of the neck (Figure 1). On palpation, the swelling was non-pulsatile; local temperature was not elevated; and it was soft and mobile with

normal overlying skin.

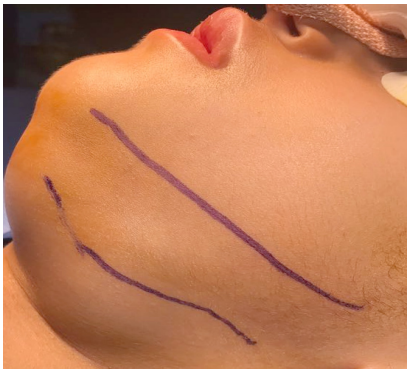


Figure 1: Lateral view of a smooth, non-tender cystic mass measuring approximately 7.0 cm × 5.0 cm was noted just below the jawline in the left lateral part of the neck.

Diagnostics

The workup included a Complete Blood Cell Count (CBC) with differential and a C-reactive protein. The CBC showed slightly elevated platelets and normal leukocytes, indicating acute-phase inflammation. The C-reactive protein was minimally elevated. An MRI with contrast was performed. The MRI showed multilocular neck lesion anterior and superior to the left sternocleidomastoid muscle with enhancing tissue extending to the submucosal left tonsillar fossa. The lesion consisted of multilocular, homogenous masses (8.0 cm x 5.0 cm in maximum diameter), which were consistent with cystic lesions (Figure 2).

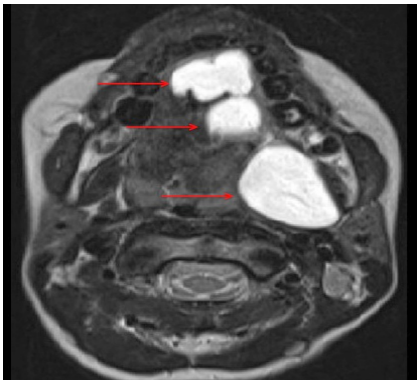


Figure 2: MRI with contrast showed a left multilocular neck lesion anterior and superior to the sternocleidomastoid muscle with enhancing tissue extending to the submucosal left tonsillar fossa. The lesion consisted of multilocular (red arrows), homogenous masses (8.0 cm x 5.0 cm in maximum diameter), which were consistent with cystic lesions.

Management

Complete surgical removal of the multilocular cyst and all offending epithelium was performed under general anesthesia, preserving the surrounding blood vessels and nerves. The incision was placed along Langer lines, directly over the mid-point of the cyst to achieve optimum cosmetic results (Figure 3). Inferior and superior sub-platysmal flaps were raised. The cyst was separated from the superficial layer of the deep cervical fascia that envelopes the SCM. Superficial jugular veins and cutaneous nerves were transected. The cyst was then dissected off the SCM, preserving the spinal accessory nerve. The cyst was dissected medially, carefully avoiding the carotid sheath and Ansa hypoglossi. The cyst was dissected off the posterior belly of the digastric and stylohyoid muscles. The rest of the multilocular cyst was fully excised trans-orally via an incision in the floor of the mouth. The wound was irrigated and closed in multiple layers, including the superficial layer of the deep cervical fascia. A regular follow up, of 6 months, showed no evidence of re currency.

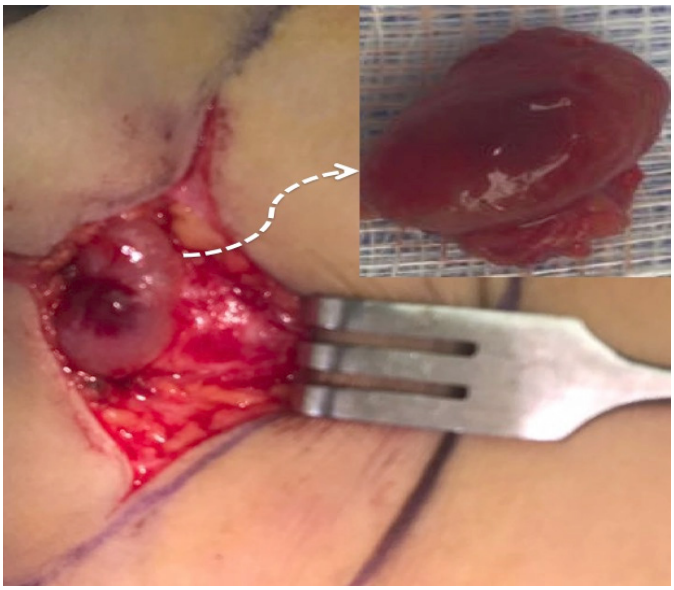


Figure 3: Complete surgical removal of the multilocular cyst and all offending epithelium was performed extra / intra-orally, preserving the surrounding blood vessels and nerves. The incision was placed along Langer lines, directly over the mid-point of the cyst to achieve optimum cosmetic results. The inset highlights the surgical excision of one entity of the multilocular cystic lesion.

Histopathology

Histopathology study showed irregular cystic structure in the dermis. The cyst was lined by two different epithelia. Outer stratified squamous and inner columnar ciliated epithelium with peripheral cells being cuboidal. There was mild inflammatory infiltrate of lymphocytes in the stroma (Figure 4). The lining cells were seen to be producing mucin in foci. The features were suggestive of BCC. The histopathology differentiated it from mucus retention cyst as mucus retention cyst shows several spaces within the connective tissue filled with mucinous material, but without an epithelial lining. Thus, clinical as well as histopathological findings confirmed the diagnosis of second BCC.

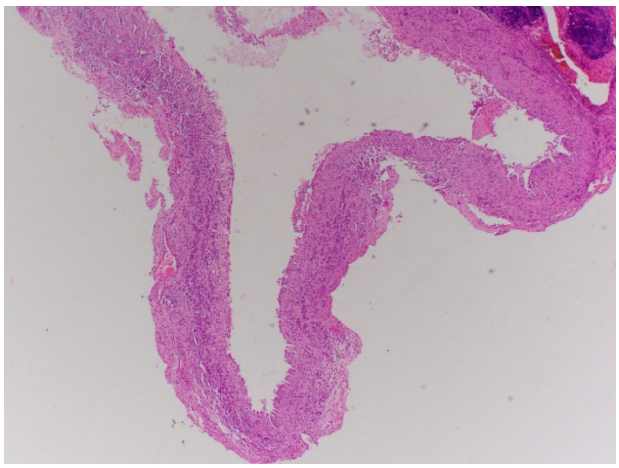
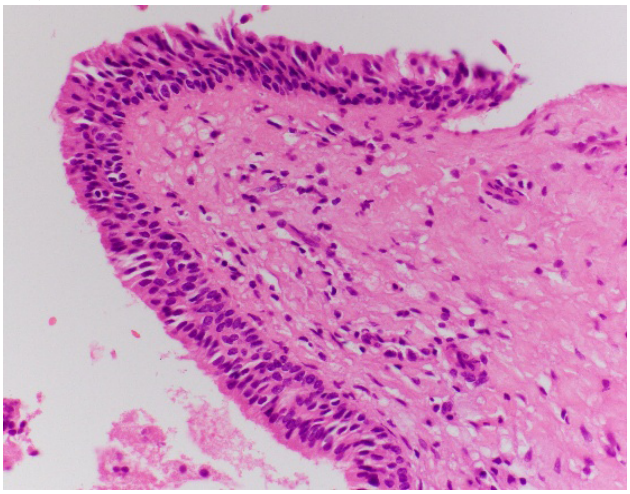


Figure 4: A) Cyst wall seen as fibro-collagenous flat tissue fragment (HEx40).



B) Inset highlights the two-different epithelium lining. Outer stratified squamous and inner columnar ciliated epithelium and presence of chronic inflammatory cells (HEx200). The lining cells were seen to be producing mucin in foci. The features were suggestive of branchial cleft cyst.

Discussion

The clinical differential diagnosis of BCC includes odontogenic infection, parotid swelling, tuberculous lymphadenitis, lipoma, cystic hygroma, carotid body tumours, thyroglossal duct cysts, suppurative lymphadenitis, branchial fistulas or sinus, dermoid cysts, neurofibroma, haemangioma, lymphangioma, teratoma, ectopic salivary tissue, pharyngeal diverticulum, laryngocele and plunging ranula. Clinicians must consider malignancies involving the lymph nodes, either primarily or secondarily, especially when the patient is elderly [2,7-9]. The cyst in this case showed a positive transillumination test; however, it is not brilliantly positive which, along with clinical history, excludes the possibility of cystic hygroma. The positive trans-illumination test and the fluctuant nature of the swelling also rules out the solid masses such as tumours involving various tissues in the submandibular region and neck as well as cervical lymphadenopathies. Besides, pharyngeal diverticulum and external laryngocele manifest in middle or old age and so will not be considered in the differential diagnosis for this case. Plunging ranula is located in the submandibular region and extends downwards.

Thyroglossal duct cysts present as a midline neck mass at or below the level of the hyoid bone and the mass moves with swallowing. Some patients will have neck or throat pain or dysphagia. Dermoid cysts occur in the midline and differ in location from the cyst in the present case. All of these differ in microscopic features from BCCs. Thymic cysts and parathyroid cysts may be included in the differential diagnosis, especially on CT evaluation. Thymic cysts occur as soft-tissue swellings in the region from the level of hyoid bone up to the mediastinum. Similarly, parathyroid cysts are located around or are most commonly inferior to the thyroid gland. However, both can be differentiated from the BCC by their typical histopathological features. As seen clinically and via MRI examination the cyst in this case was located above the clavicle and along a path between the anterior margin of the sternocleidomastoid muscle, and the tonsillar fossa which is a characteristic of a second BCC [2,9-11]. Surgical excision of the BCC is the treatment of choice and is considered definitive. Complications of surgical treatment include recurrence, formation of a persistent fistula and damage to the cranial nerves.

Alternative treatments, such as percutaneous sclerotherapy, are promising but remain unproven [8,11]. Recurrences are known to occur following complete surgical excision of the BCCs, as seen in a large retrospective study where the overall recurrence rate was noted to be 4.9% after a 2-year follow-up period, while in other studies a recurrence rate of 3-20% was reported [5,9,10]. In this study there was no recurrence on a regular six months follow up. In conclusion, the aetiology of BCC is yet to be determined, although the majority of opinions suggest its origin is either from branchial apparatus or from lymphoid tissues. Considering the anatomical location and the radiological appearance, the precise embryological origin can be predicted. It seems to be that this is the first case to be reported of a multilocular branchial cleft cyst, excised of this size, from the oropharyngeal area in a child. BCC shares a clinical presentation with other pathological entities of the neck, making diagnosis difficult at times. Oral health professionals should be aware of this entity because it can be easily confused with an odontogenic infection and submandibular or parotid gland pathology, especially if it develops high up in the neck.

Declaration

The author declares no conflict of interest. The author has no financial interest in this study. The study received no financial fund by any organization and / or institute

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