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Case Report

Submental midline branchial cleft cyst: a first case report in literature

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ABSTRACT

A branchial cleft cyst is a congenital abnormality typically located over the lateral aspect of neck. A 3 year old boy presented with a gradually progressive painless swelling below his chin since 1 year. CT scan demonstrated well defined cystic lesion in submental region. Excision was done and histopathology showed the cyst lined by stratified squamous epithelium and at places lined by pseudo stratified ciliated columnar epithelium with subepithelial lymphocytes suggesting branchial cyst. Branchial cleft cysts should also be considered as one of the differential diagnoses in cystic midline lesions of the neck, if it's not moving with deglutition and with tongue protrusion.

Keywords: Submental region, Midline, Branchial cleft cyst

INTRODUCTION

A branchial cleft cyst (BCC) is a common congenital abnormality that is seen over the lateral aspect of neck mainly along the anterior border of the sternocleidomastoid muscle, but they have also been reported rarely at uncommon locations such as nasopharynx, thymus, thyroid gland, oral cavity, salivary glands, pancreas and mediastinum.[1-8] It is also known as congenital hydrocoele of the neck, hygroma colli and lateral lympho-epithelial cyst.[2] Here we report a case of pediatric midline submental BCC and to the best of our knowledge; such a case has not been previously reported.

CASE REPORT

A 3 year old child presented with a gradually progressive painless swelling below his chin since 1 year. On clinical examination, a solitary diffuse swelling of size 5×4 cms was present in the midline of the neck occupying the submental triangle. It was not moving with deglutition and tongue protrusion. It was non-tender, soft in consistency and fluctuant (Figure 1). Ultrasonography showed a well defined thick walled cystic lesion in the submental region with internal echoes within the cyst, suggestive of dermoid cyst. CECT neck showed a well defined hypodense, non-enhancing cystic lesion in the left sublingual space measuring 4.1×2.1×3.9 cms. It extended superiorly compressing the left genioglossus and hyoglossus, inferiorly reaching up to the hyoid bone without eroding & posteriorly compressing the left submandibular gland and reaching till the base of tongue (Figure 2). FNAC was not done as the child wasn't co-operative for the procedure. Based on these clinical and investigative findings, a provisional diagnosis of dermoid cyst was made and excision was done (Figure 3). Histopathology revealed a fibrocollagenous cyst wall lined by stratified squamous epithelium and denuded at places which was lined by pseudo stratified ciliated columnar epithelium and replaced by macrophages arranged in sheets. Amidst macrophages, multinucleated giant cells were seen. The cyst wall showed adnexal structures and chronic inflammatory infiltrates consisting of lymphocytes and plasma cells in the sub epithelial layer (Figure 4). The postoperative period was uneventful and child was in regular follow up for 6 months.

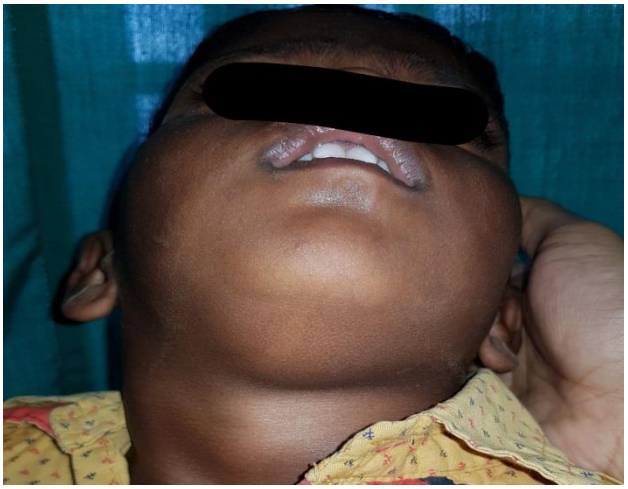

Figure 1: Midline submental swelling of a 3 year old boy.

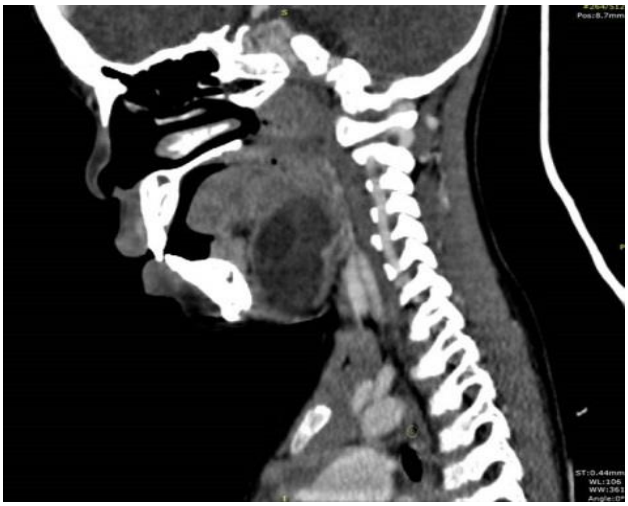

Figure 2: CECT showing well defined, hypodense, non-enhancing cystic lesion in the left sublingual space.

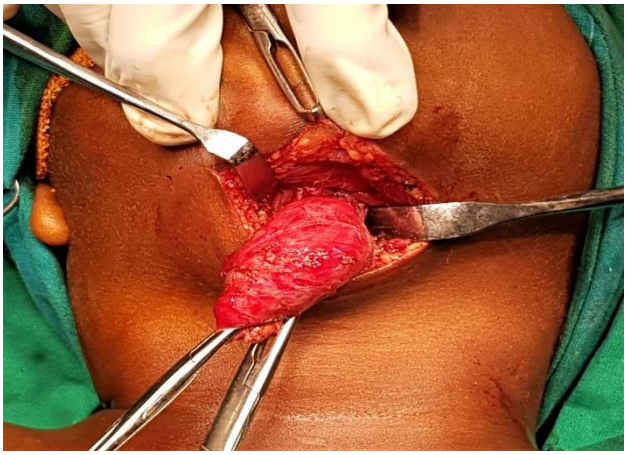

Figure 3: Intra-operative picture of the swelling.

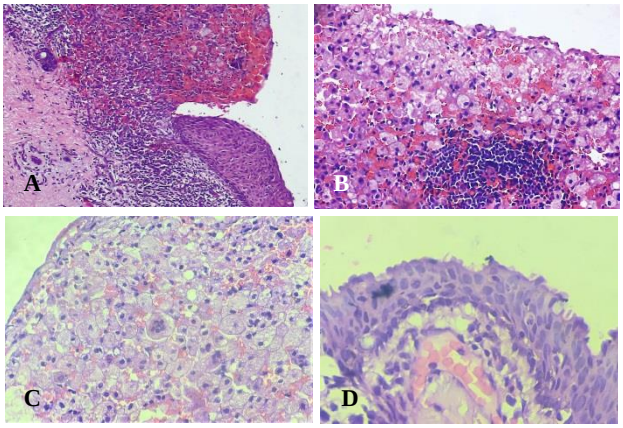

Figure 4: (A) Cystic lesion lined by stratified squamous epithelium with chronic inflammation in the wall; (B) denuded epithelium replaced with sheets of macrophages; (C) multinucleated giant cells are seen amidst macrophages; (D) the lining epithelium at places shows pseudostratified ciliated columnar epithelium.

DISCUSSION

BCCs are the most common type of lateral congenital neck swellings. Around 30% of congenital neck swellings are BCCs. Among all the branchial arch anomalies in children, fistulas are common followed by sinuses and cysts. 95% of the BCCs originate from second arch and the remaining 5% from first, third and fourth arches.[1, 2] Apart from these normal sites, there are unusual locations of BCC cases reported in the literature. The other sites are thyroid, nasopharynx, oral cavity, mediastinum, thymus and pancreas. Chung et al described an extremely rare case of bilateral intrathyroidal branchial cleft cysts in a 2 day old baby.[4] Aggarwal et al reported a case of BCC in the midline of neck just below the hyoid bone.[9] Kumara et al in their study reported a rare case of BCC in the floor of the mouth.[5] After extensive literature search, this was the first case presented in the submental midline region.

There are various theories which explain the origin of branchial cyst i.e., incomplete obliteration of branchial mucosa, cervical sinus of His theory, thymo-pharyngeal duct theory and cystic degeneration of lymph node.[2] According to King, any cyst in the lateral wall of neck with lymphoepithelial features is regarded as branchial cyst.[1,10] Our case is an exception for King's criteria as this was in the midline submental region.

Ultrasonography of BCC shows a well defined, anechoic thick walled cystic lesion with posterior acoustic enhancement in most of the cases.[11,12] In our case there was a well defined cystic lesion with posterior acoustic enhancement. BCCs on CT scan shows well defined, low attenuated, non-enhancing cystic lesion with uniform thickness.[12] The wall thickness and enhancement will vary depending on the severity of the associated

inflammatory process within the cyst. Occasionally the cysts are hyper dense due to intra cyst hemorrhage from FNAC. MRI is needed to assess the deeper extent of the lesion. The cyst shows hypo to intermediate signal intensity on T-1-weighted scans and hyper intense on T-2-weighted scans.[12] The other differential diagnoses in this region include lymph node enlargement, thyroglossal duct cyst and dermoid cyst. A dermoid cyst will appear as thin walled, well defined, hypo attenuating; unilocular masses on CT scan which is similar in appearance to branchial cyst.[13] Thyroglossal duct cyst are well circumscribed anechoic cystic lesion in the midline but are related to the undersurface of hyoid bone.[4] In our case we had diagnosed it as a dermoid cyst but the histopathology turned out to be BCC.

Histopathologically, the lining of the cyst wall is by stratified squamous epithelium but at some places the cyst wall is denuded and replaced with pseudostratified ciliated columnar epithelium. Sub epithelial layer has chronic inflammatory cells, lymphocytes and plasma cells organized in germinal centres.[2] The germinal center organization was not there in our case. The absence of adnexal structures in the cyst wall distinguishes it from dermoid cyst. Thyroglossal duct cysts have nonkeratinizing stratified epithelium with presence of thyroid follicles in the cyst wall which differentiates it from the present case.[4] Surgery is the mainstay of treatment for BCC. Sclerotherapy is the other option available for lesions not amenable for surgery. Picibanil (OK-432) is the most commonly used sclerosing agent.[14] According to Roh et al there was complete disappearance of the BCC in 58% of cases if injected for one to three times.[14]

CONCLUSION

BCC is a commonly occurring developmental cyst in the lateral neck region. But when compared to its usual presentations, this case was an exception, as it was found in the midline submental region. Hence, BCC should also be kept in mind as one of the differential diagnoses in cystic midline lesions of the neck, if it's not moving with deglutition and with tongue protrusion.

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