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Cystic Lymphangioma Of The Right Parotid Gland In An Infant: A Rare Entity With Classic Imaging Features

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ABSTRACT

Cystic lymphangioma is a benign lesion consisting of multiple lymphatic spaces. The primary site of lymphangioma at parotid gland is extremely unusual and till date, there are only very few cases of infant cystic lymphangioma of the parotid. In this study, we presented the case of a 7-month-old female patient with cystic lymphangioma of the right parotid gland since birth which is a rare entity and its classic imaging features in detail. USG revealed a well-defined multiloculated, multiseptated anechoic cystic lesion involving the right parotid gland, likely involving both superficial and deep lobes. The lesion measured approximately 45 x 40 × 31 mm (CC x AP × TR). Inferiorly, the lesion extended into the right submandibular region, causing mass effect on the ipsilateral submandibular gland, which was displaced anteromedially. CECT showed well-defined multilobulated, multiseptated hypodense cystic lesion with attenuation values of 10–20 HU involving the right parotid gland, measuring approximately 48 × 43 × 35 mm (CC × AP × TR). The lesion demonstrated thin, imperceptible walls and internal septations. As per this study, USG can be recommended for initial non-invasive imaging that leads to a diagnosis. To evaluate the extent of the lesion, CECT had been used in this study which provided better diagnosis. However, the definitive diagnosis can be achieved with final histopathological examination.

1. INTRODUCTION

Lymphangiomas are benign lymphatic malformations most frequently observed in the young age group and, uncommonly, in the elderly population[1]. Cystic lymphangioma is a benign lesion consisting of multiple lymphatic spaces[2]. It is uncommon congenital anomaly and usually present as an asymptomatic, painless, soft, fluctuant mass in infancy, and early childhood. Their occurrence is due to an embryonic developmental anomaly of the lymphatic system[3]. The head and neck are the most common locations for lymphangioma to occur. Other less common sites are the axilla, shoulder, chest and abdominal wall[4]. The primary site of lymphangioma at parotid gland is extremely unusual and till date, there are only very few cases of infant cystic lymphangioma of the parotid. In this study, we presented the case of a 7-year-old female patient with cystic lymphangioma of the right parotid gland since birth which is a rare entity and its classic imaging features in detail.

2. CASE REPORT

A 7 months old female child presented with a progressively enlarging, painless swelling in the right parotid region since birth. Figure 1 showed photograph with visible swelling over the right side of the cheek and upper neck region, corresponding to the anatomical location of the right parotid gland. Over the past few days, the swelling had increased in size and was associated with intermittent fever.

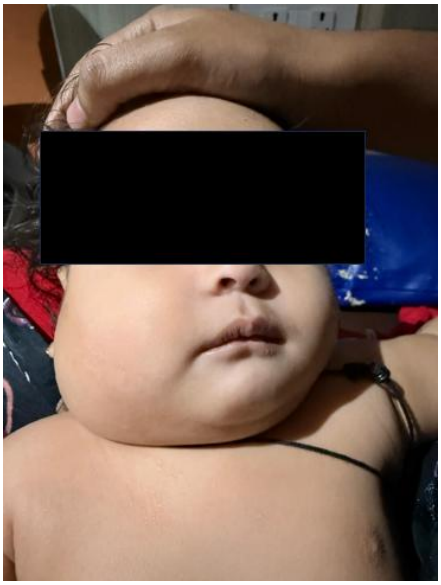


Figure 1. Clinical presentation of cystic lymphangioma

Birth history: Full-term infant delivered via lower segment cesarean section (LSCS) due to cord around the neck; birth weight 3.5 kg. No NICU admission was required. Immunization was up-to-date till 3.5 months of age, and developmental milestones were appropriate.

Ultrasound Examination was advised and revealed a well-defined multiloculated, multiseptated anechoic cystic lesion involving the right parotid gland, likely involving both superficial and deep lobes. No internal echoes, solid components, or vascularity were seen. The lesion measured approximately 45 x 40 × 31 mm (CC x AP × TR), with the largest locule measuring 12 × 37 mm and internal septae up to 2.4 mm thick. Inferiorly, the lesion extended into the right submandibular region, causing mass effect on the ipsilateral submandibular gland, which was displaced anteromedially.

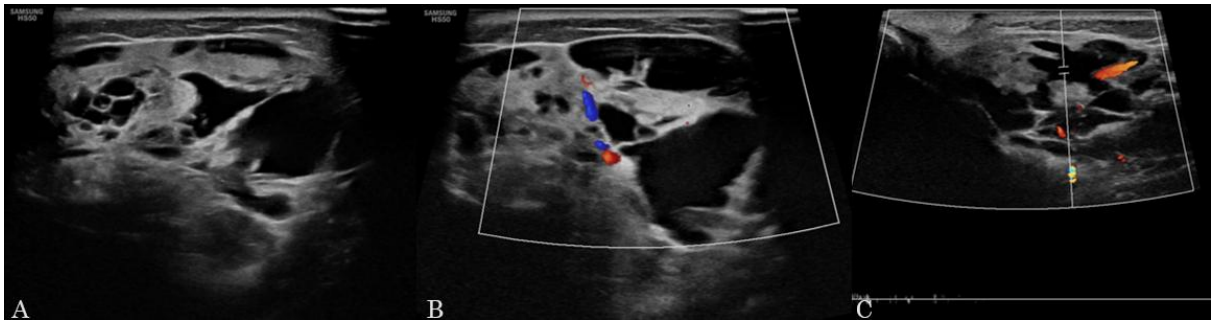
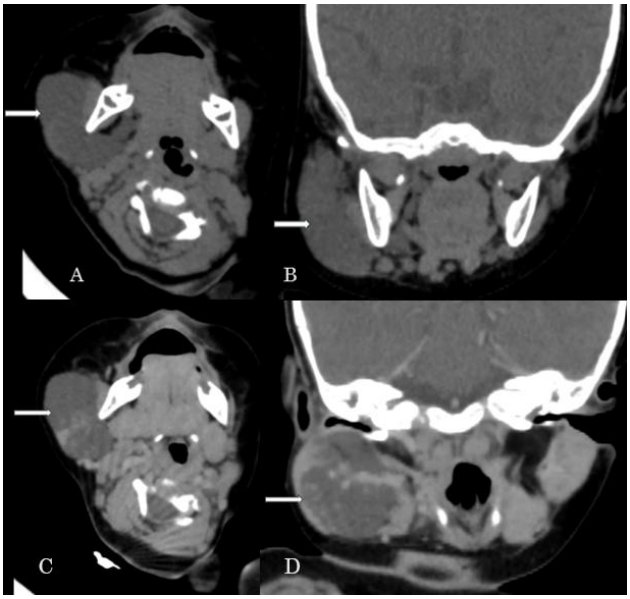


Figure 2 A: Gray-scale ultrasound image demonstrating a multiloculated anechoic cystic mass involving the right parotid gland. The lesion appears well-defined with thin internal septations and no solid components. Figure 2 B: Color Doppler ultrasound image of the same lesion shows absence of internal vascularity, with no flow signals appreciated within the cystic areas or septations. Figure 2 C: Spectral Doppler tracing reveals no detectable waveform within the lesion, further supporting the avascular nature of the cystic mass.

Further CECT neck was advised. It revealed a well-defined multilobulated, multiseptated hypodense cystic lesion with attenuation values of 10–20 HU involving the right parotid gland, measuring approximately 48 × 43 × 35 mm (CC × AP × TR). The lesion demonstrated thin, imperceptible walls and internal septations showing mild post-contrast enhancement, while the rest of the lesion remained non-enhancing. Interspersed enhancing parotid parenchyma was noted. Medially the lesion was seen abutting the right submandibular gland. Anteromedially the lesion is in contact with the ramus and condylar process of the right hemimandible, without bony erosion. Posteromedially the lesion is in contact with the right internal jugular vein and sternocleidomastoid muscle. Superiorly it abuts the right external auditory canal. Other visualized structures including the nasopharynx, oropharynx, larynx, trachea, esophagus, thyroid, left salivary glands, and cervical spine were unremarkable. Above findings are suggestive of cystic lymphangioma of the right parotid gland.



Figures 3 A and 3 B: Axial (A) and coronal reformatted (B) non-contrast CT images demonstrate a well-defined, multilobulated, multiseptated hypodense cystic lesion in the right parotid gland (arrows), with an attenuation range of 10–20 Hounsfield units. Figures 3 C and 3 D: Axial (C) and coronal reformatted (D) post-contrast CT images reveal no significant contrast enhancement of the lesion. The thin, imperceptible cyst wall and internal septae show mild post-contrast enhancement (arrows).

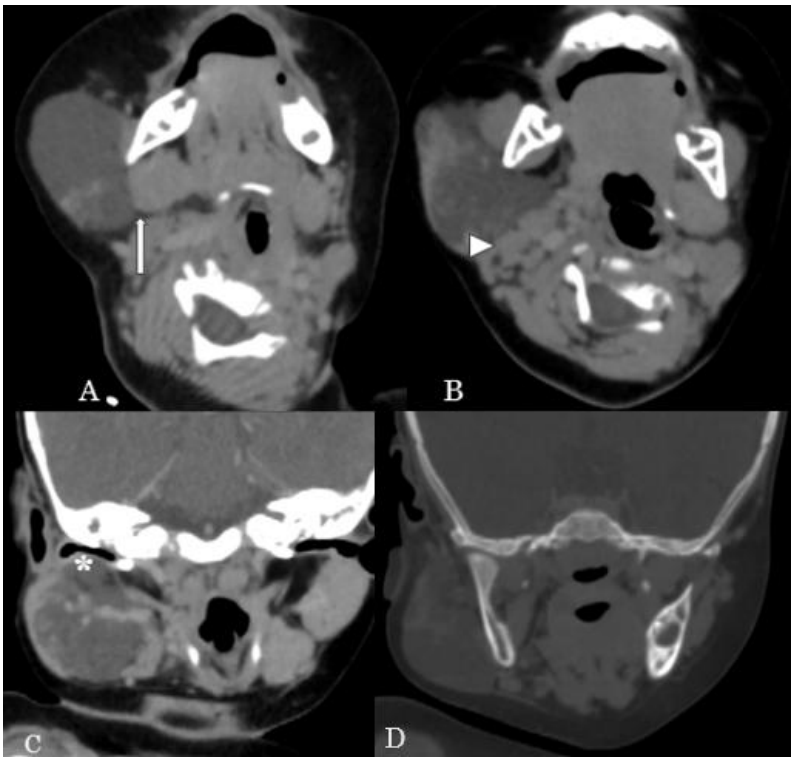


Figure 4 A: Axial CT image shows the lesion abutting the right submandibular gland medially (arrow). Figure 4 B: Coronal reformatted CT image demonstrates postero-medial abutment of the lesion with the right internal jugular vein and the sternocleidomastoid muscle (arrowhead). Figure 4 C: Axial CT image reveals superior contact of the lesion with the right external auditory canal (*). Figure 4 D: Bone window axial CT image shows the lesion abutting the ramus and condylar process of the right hemimandible, without evidence of bony erosion.

3. DISCUSSION

Lymphangiomas are developmental anomalies rather than true neoplasm. Embryologically, the lymphatic system arises from 6 primitive sacs that develop at the 6th week of intrauterine life. The first pair is jugular sac, the second pair is cisterna chyli

of retroperitoneal tissues, and the third pair is posterior lymph sacs which develop at the inguinal region. These lymphatics of various regions develop communication with each other and with the venous system. It is established that the majority of lymphangiomas arise from parts of lymph sacs that fail to make communication. The disconnected lymphatic structure may dilate and evolve to a cystic formation. Its aberrant proliferation led to cystic lymphangioma with a common presentation as large soft-tissue mass[4, 5].

Depending on the histomorphology, lymphangiomas are classified as lymphangioma simplex, cavernous lymphangioma, and Cystic Lymphangioma or cystic hygroma. Cystic types have the potential for extensive infiltration of surrounding tissues and lead to surgical difficulties. Therapeutically, they are classified into macrocystic, microcystic or mixed lymphangiomas[6].

Cystic lymphangioma within the parotid are uncommon, and clinically, they are misdiagnosed as tumors. During dealing with cystic lesions of the parotid gland, they can be misdiagnosed as neoplastic lesions such as cystic pleomorphic adenoma, cystic low-grade mucoepidermoid carcinoma, Warthin tumor, acinic cell carcinoma, and non-neoplastic lesions such as retention cyst, branchial cleft cyst and polycystic disease of the parotid gland[7,8].

Clinically, cystic lymphangioma usually present as a painless, soft, cystic swelling that enlarges over time. Fluctuation and transillumination of the parotid gland showed positive in cases of lymphangioma. The cyst may present for a long duration without any symptoms. Complications of the cyst may include the infection or hemorrhage leading to the effects of compression on the facial nerve and cystic rupture. There were no reported cases of malignancy associated with cystic lymphangioma[5].

Proper history, clinical examination, and imaging studies play an important role to confirm the diagnosis of cystic lymphangioma and to exclude other possible differentials of congenital lesions. Computed tomography helps to delineate extension and anatomy study before operation to avoid iatrogenic injury to vital structures. As per Gold et al, USG of a lymphangioma typically show a multicystic, hypoechoic lesion with thin walls which is in accordance with our USG report of case included[9]. A study by Mandel et al[10] showed computed tomography (CT) finding of a homogeneous cystic lesion with a surrounding smooth, thin wall which showed similar findings like our study. CT can also show the involvement of the deep lobe and enhancing pattern of the cyst.

The treatment usually involve the complete excision of the cyst. however, if inadequate excision occurs the rate of recurrence ranges from 10% to 38%. Surgical options to consider are enucleation and superficial or total parotidectomy. For patients with a macrocystic lesion unwilling to undergo surgery, sclerosing agents such as OKT-3 and bleomycin may be used with varying results. Intralesional injection with such agents has a role in the medical management of these cases. The use of radiation therapy has no proven role in the management of lymphangioma and adds the risk of malignant transformation[11].

4. CONCLUSION

Cystic Lymphangioma is an uncommon congenital lymphatic lesion that uncommonly affects parotid gland. Most patients are asymptomatic. When symptoms do occur, the lymphangioma requires intervention. As per this study, USG can be recommended for initial non-invasive imaging that leads to a diagnosis. To evaluate the extent of the lesion, CECT had been used in this study which provided better diagnosis. However, the definitive diagnosis can be achieved with final histopathpological examination.

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