A case report: Neonatal infected congenital sublingual cyst with upper airway obstruction.

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Abstrast

Congenital sublingual cyst (CSC) is an exceptionally rare anomaly in newborn, with limited case studies published to date. The occurrence of symptomatic CSC in newborn is extremely uncommon, and this case is the first reported case of a neonate with an infected CSC leading to airway obstruction.

A three-week-old female infant presented at the emergency room with symptoms of aspiration pneumonia and upper airway obstruction attributed to a substantial turbid within sublingual cyst. Emergency measures included securing her airway by Dual PAP and admitted to NICU. Immediate bedside intervention involved needle aspiration, revealing a cystic content comprising 18 ml of pus. Over the subsequent two weeks, the sublingual cyst gradually returned to its previous size. Consequently, a scheduled marsupialization was performed. The postoperative outcome was favorable, with successful reversal, extubation on the first day postoperatively, resuming normal oral feeding, and mild sublingual bulging. The infant was discharged on the third week, and notably, there was no recurrence of the cyst during the 18 months of follow up.

In conclusion, this reported case highlights the rare occurrence of symptomatic CSC in a newborn necessitating definitive surgical management. The goal of surgical intervention is to restore normal function while minimized the risk of recurrence and less of morbidity. This extraordinary case report serves as a valuable contribution to medical knowledge, motivation the sharing of this unique clinical experience with the broader medical community.

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Introduction

Congenital tumor's incidence is in the range of 1.7-13.5 per 100,000 live births,[1] congenital sublingual cyst (CSC) stand out as an exceptionally rare anomaly in newborns, with only limited case studies have been published. Typically, diagnosis of CSC occurs in late childhood, as these cysts are often asymptomatic at birth and may grow to a size that poses airway or feeding problem.[2,3] Symptomatic CSC in newborn is extremely uncommon, necessitation a surgical management. Radiological assessments play a crucial role in establishing the diagnosis by precisely locating the cyst and determining its content.[2,4]

The optimal treatment remains controversial. Some advocate for early surgical intervention to prevent complications, while others propose a watchful observation strategy with surgery reserved for case with evident airway or feeding issues.[5-7] The goal is to restore normal function and minimize the risk of recurrence with less morbidity.

This report highlights a unique case involving an infected CSC in a newborn, leading to airway compromise. The emergent need for a secured airway prompted a surgical intervention involving marsupialization, a procedure not yet documented in the existing literature. This case shows aspect of CSC management, emphasizing the importance of adapting surgical strategies to address specific clinical challenges and improve overall outcomes.

Case report

A female infant born by vaginal delivery at 40weeks gestation, weighing 3,150 g., Apgar scores were 9 and 10 at 1and 5 minutes, respectively. The mother had an unremarkable medical and obstetric history, and there was no family history of any congenital anomalies. On clinical examination the infant exhibited a slight elevation of the tongue, accompanied by a cyst bulge on the anterior floor of mouth, about 1.2 cm. in size. Despite this, there were no apparent issues with breathing or feeding, and the overall general examination was normal. The doctor recommended referral to an excellent center for further evaluated as outpatient, but financial constraints prevented the mother from pursuing this option.

At the three weeks of age, the infant presented at the emergency room with a fever, increased difficulty breathing while in a supine position, and poor feeding over the preceding two days. Physical examination was BT 38 C, RR 64 bpm, PR 122 bpm, dry lip, and poor air entry in the lung with subcostal

retractions. A diagnosis of aspiration pneumonia with upper

airway obstruction was made, and the airway was secured by DuoPAP and admitted to NICU. Further examination revaluated the sublingual cyst, which appeared more enhanced, turbid and fluctuant (Figure 1). The differential diagnosis included an infected ranula cyst, dermoid cyst, thyroglossal duct cyst, and lymphangioma. Bedside aspiration yielded 18 ml of pus, which was sent for gram strain and bacterial culture (figure 2). Treatment included intravenous Amoxicillin/clavulanic acid, intravenous fluid and a retained oral gastric tube for feeding.

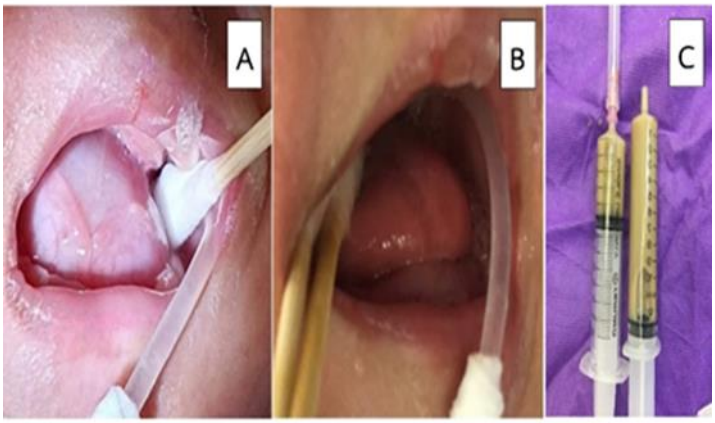

Figure 1. Clinical pictures of the treated patient.

A. A well-defined, symmetrical sublingual swelling was found, with no inflammatory signs, covered by normal-looking mucosa and about 3.2 cm in diameter. Upon palpation, the lesion showed a soft consistency with fluctuation. Due to the volume of the lesion, the tongue was raised and displaced posteriorly, no any discharge per Wharton's duct.
B. Post bedside aspiration for symptomatic relief and content investigation, a clinical examination of the oral cavity revealed a downward position of the tongue, and there was mild swelling observed in the sublingual region.
C. An 18-gauge needle was utilized for bedside aspiration, resulting in the extraction of aspirated content with a yellowish and viscous appearance.

Laboratory reports indicated patchy lung infiltration on chest x-ray and a hypoechogenic cystic lesion measuring 1.8x2.2x1.9 cm in the submental region, there was no gross calcification or hypervascularity, and the thyroid gland appeared unremarkable on ultrasonography. Pus culture identified *S. aureus* sensitive to Amoxicillin/clavulanic acid, while electrolyte and hemoculture showed no abnormalities.

After a week, the infant still had a low-grade fever, mild sublingual bulging, and successfully to transitioned off ventilator support, moving to the pediatric ward for continuous intravenous antibiotics which changed to Vancomycin to cover hospital-acquired *S. aureus* and Amikacin to cover gram-negative hospital-acquired infections.

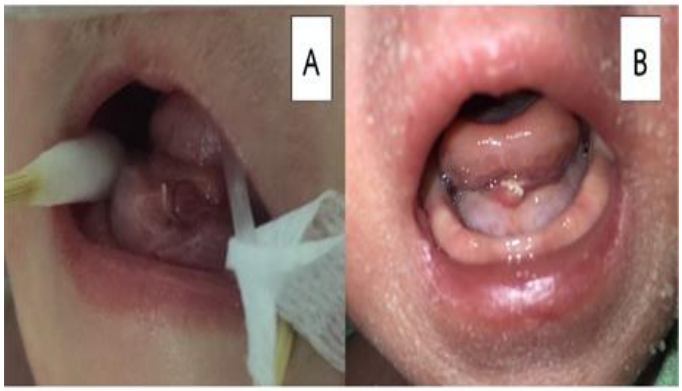

Figure 2. After marsupialization.
A. Post-op Day 1: mild swelling of sublingual region and the tongue was slightly raised. Absorbable suture of edge of wound with oral mucosal.
B. Post-op Day 7: the floor of mouth exhibited good health, and the tongue had returned to its normal position.

Two weeks later, despite the persisting low-grade fever, the sublingual cyst gradually increased in size. Consequently, marsupialization was scheduled. The cyst yielded 15 ml of pus, which was sent for gram strain and culture. The infant recovered successfully, was off the ventilator on the first postoperative day, normal oral feeding, displayed mild sublingual bulging (Figure 2), and was discharged on the third week. Over the subsequent 18 months of follow-up, there was no recurrence of the cyst, and the infant exhibited normal development in feeding and speech.

Discussion

The CSC in newborn is exceptionally rare, with limited case studies documented in the literature. Typically, the diagnosis of CSC occurs in late childhood, as these cysts often go unnoticed at birth and subsequently grow, potentially causing airway or feeding issues.[2,3] Only two cases of symptomatic CSC in newborn were described in the literature.[8,9] This is the first reported case of a neonatal infected CSC with airway obstruction, where need needle aspiration

failed, and successful marsupialization was employed.

The patient presented with airway compromise and feeding difficulties associated with a sizable sublingual cyst. The physical examination of lesion showed turbid, well-circumscribed and fluctuated whose differential diagnosis, including ranula cyst, dermoid cysts, epidermal cyst, lymphatic malformation, vascular malformation, teratoma, thymoma, and ectopic thyroglossal duct cyst.[2,4] Radiologic workup may help establish the diagnosis, in this case's ultrasonography showed hypoechoic cyst at the submental region which no gross calcification or hypervascularity, unremarkable of thyroid gland. While the clinical and ultrasonography finding favored ranula cyst, other possibilities to considered such as dermoid or epidermoid cyst which appear as cystic lesions in the midline of sublingual or submental spaced[4] and less likely possibility for solid-cystic lesion (lymphatic malformation, teratoma).[2]

Ranula cyst, characterized by the dilatation of a sublingual gland, is localized anteriorly on the floor of the mouth between the mylohyoid and hyoglossus muscles, elevating the tongue.[2] The cause of ranula cyst in newborn remains unknown, potentially arising from a congenital failure of canalization of the terminal end of the duct.[7] Other hypotheses may be more plausible accounting for the increased incidence in sublingual glands[9]. The definite diagnosis of congenital ranula cyst is often confirmed through histology, although it was not performed in this case.

The treatment protocol for sublingual cyst in newborn remains controversial. Options of treatment include observation for spontaneous resolution in uncomplicated cases, early surgery for preventing complications and surgical management to relieve symptoms.[2,5-7,9-10] In this case, with airway compromise and feeding difficulties, definitive surgical management was necessary. Needle aspiration was initially attempted due to its bedside, quick, easy, less complication, but recurrence occurred in this instance. Other surgical treatment options are marsupialization, excision, cryosurgery, and sclerotherapy.[2,7] Marsupialization was chosen as the second surgical intervention for this case due to its advantages of shorter general anesthesia time, low risk of complications such as Wharton's duct injury, lingual nerve paraesthesia, bleeding, or infection, despite a recurrence rate of 66.7%.[11]

In conclusion, symptomatic sublingual cyst is uncommon in newborn, and surgical intervention becomes necessary to restore normal function and reduce the risk of

recurrence with minimal morbidity. This case report is extraordinary and motivated to share the clinical experience to the medical community**.**

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Ethical approval: written consent was obtained from the legal representative of patient (Her parents) for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor.

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