

CASE REPORT OPEN ACCESS

Sublingual Epidermoid Cyst in a Neonate—A Rare Case Report on Early Detection and Intervention

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ABSTRACT

Epidermoid cysts are rare benign developmental lesions that occur infrequently in the oral cavity, accounting for less than 0.01% of oral cysts. Congenital sublingual epidermoid cysts in neonates are extremely uncommon but may interfere with feeding and tongue movement. This report presents a rare case of a congenital sublingual epidermoid cyst in an 11-day-old neonate managed successfully with early surgical excision to restore normal feeding function. An 11-day-old male neonate from Ethiopia presented with a congenital swelling on the floor of the mouth beneath the left side of the tongue, which was noticed immediately after birth. The swelling, measuring 4 × 3 cm, was noticed immediately after birth. The initial fine needle aspiration cytology (FNAC) revealed a sublingual keratin cyst. Clinically, epidermoid cysts are characterized by a rubbery consistency on palpation, well-defined borders, and varying sizes. They are usually asymptomatic and slow-growing. The final diagnosis is usually confirmed by histopathological examination. Surgical excision, with the goal of complete removal including the cyst wall, is the definitive treatment option to prevent recurrence. If left untreated, the lesion may grow significantly and lead to complications such as dysphagia, breathing difficulties, pain, and reduced tongue movement.

1 | Introduction

Congenital epidermoid cysts are rare, benign, usually asymptomatic, and slowly growing lesions arising from ectodermal tissue. Congenital cysts of the oral cavity are uncommon conditions [1]. It may occur due to retained epithelial remnants in the midline during the closure of first and second brachial during the third and fourth weeks of intra uterine life [2]. Epidermoid cysts can occur throughout the body and represent approximately 7% of cysts in the head and neck region and 1.6% of those in the oral cavity [3]. Although rare in the oral cavity, most occur in the midline of the sublingual region. Epidermoid cysts may occur on the skin, mucous membranes, and also visceral organs. The epithelial lining of cysts produces keratin [4]. Ultrasound is the first imaging modality used to diagnose sublingual epidermoid

cysts due its reliability and cost-effectiveness [5]. This case report presents a rare sublingual epidermoid cyst in an 11-day-old male child. Early surgical intervention was performed for better feeding.

2 | Case History/Examination

An 11-day-old male neonate from Ethiopia presented with a congenital swelling on the floor of the mouth beneath the left side of the tongue that was noticed immediately after birth. The swelling was noticed immediately, prompting close observation in the pediatrics department. A Naso Gastric (NG) tube was placed due to difficulty suckling. He was then referred to the Oral and Maxillofacial surgery department. Upon physical examination,

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Key Clinical Message

Congenital sublingual epidermoid cysts are extremely rare in neonates but may cause feeding difficulty and tongue displacement. Early diagnosis using clinical examination and imaging followed by complete surgical excision is essential to prevent airway compromise and feeding problems, allowing recovery and normal oral function in newborn infants.

a well-circumscribed, solitary, fluctuant, and non-tender mass measuring 4×3 cm was found in the left sublingual area. The mucosa overlying the lesion appears normal in color and texture. There was significant deviation of the tongue toward the right side, with a slight elevation and limited movement (Figure 1). On extraoral examination, no submental swelling and cervical lymph nodes are non-palpable.



FIGURE 1 | Left side sublingual epidermoid cyst (green arrow) clinical picture, pushing the tongue superiorly and laterally.



FIGURE 2 | (A) Completely excised epidermoid cystic mass, (B) the cystic pocket with its lining.

3 | Differential Diagnosis, Investigations and Treatments

The differential diagnosis included dermoid cyst, ranula, lymphatic malformation, and salivary gland cyst. A fine needle aspiration cytology (FNAC) was performed, suggestive of keratin cyst. The sonographic investigation revealed a well-defined, round, homogenous hypoechoic cystic content in the floor of mouth. There was no evidence of vascularization. The child was diagnosed with a congenital sublingual cyst. After obtaining normal results from other baseline investigations, the patient was taken to the OR and orally intubated. Following routine preparation and draping, a throat pack was placed, a single suture was passed through the tongue for suspension, a transverse incision was made, and careful dissection of the cyst from adjacent tissue and complete excision of the lesion with its lining was done (Figure 2A,B). Hemostasis was successfully achieved, and the mucosa was closed primarily.

4 | Outcome and Follow-Up

The patient recovered from anaesthesia and was postoperatively placed on an NG tube for 3 days. After this, the NG tube was removed, and the patient began suckling breast milk without any issue. The child's postoperative period was smooth and without any complications, with no complaints reported by the parents on follow-up appointments. Microscopic examination of the specimen showed cystic tissue with a wall lined by bland squamous epithelium with a focal area of pseudostratified columnar epithelium with underlying fibromuscular stroma with mucous glands (Figure 3A–C). The patient remained under follow-up care and was progressing well with feeding. Written informed consent was obtained from the child's mother for publication of this case report. A copy of the written consent is available for review by the Editor-in-chief of this journal on request.

5 | Discussion

An epidermoid cyst is a rare, benign, painless, slowly growing lesion that can occur anywhere on the body. It accounts for approximately 7% of cases in the head and neck regions [6]. It is one of the three congenital developmental cysts in addition to dermoid and teratoid cysts. The main difference between dermoid and epidermoid cysts is the presence of skin appendages in the former [7, 8]. In the oral cavity, dermoid cysts are more

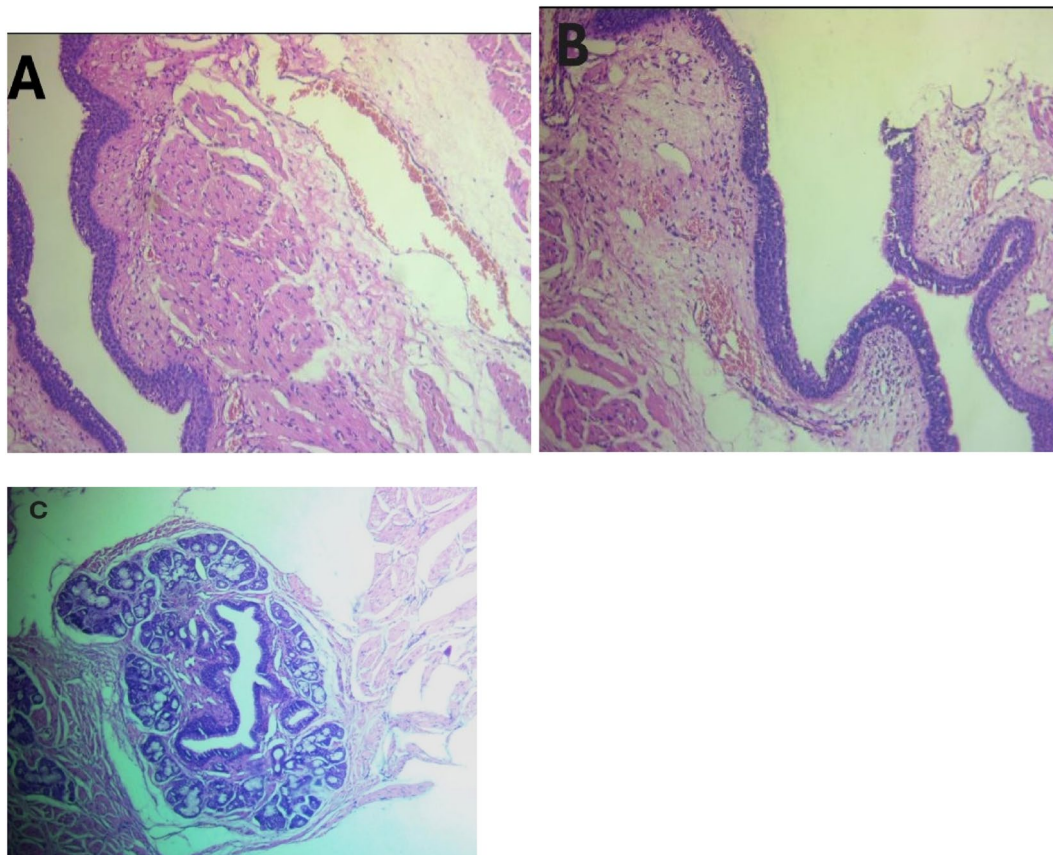


FIGURE 3 | Hematoxylin and Eosin Staining: (A, B, $\times 100$) photomicrograph showing cystic tissue with cyst wall lined by bland stratified squamous epithelium with fibrous wall. (C, $\times 200$) underlying fibrovascular stroma contains mucous glands.

common than epidermoid cysts. The occurrence of epidermoid cysts and dermoid cysts comprises only 0.01% of all the oral cavity cysts [9]. The predominant age for epidermoid cysts is between 15 and 35 years with male predilection [10]. Kumari et al. reported a sublingual epidermoid cyst in a 6-year-old child. Compared with these cases, our patient presented much earlier during the neonatal period, emphasizing the importance of early recognition and management to prevent feeding difficulty and air compromise.

Our case report describes a sublingual epidermoid cyst in an 11-day-old male child, to the best of our knowledge; very few cases have been reported in neonates, highlighting the rarity of this presentation. Epidermoid cyst can be classified into three types based on their location and relation to the muscles of the floor of the mouth: sublingual, located above the geniohyoid, also called median genioglossal cysts; geniohyoid cyst, located in the submental region between the geniohyoid and the mylohyoid muscles; and lateral cysts, which are located in the submaxillary region [11, 12]. Our current case report identifies anatomical findings in the sublingual region, classifying it as a sublingual epidermoid cyst. Clinically, the lesion may vary in size from a few millimeters to several centimeters depending on its location [13]. Patients with epidermoid cysts may experience difficulty in breathing and dysphagia, which can be caused by the upward displacement of the tongue due to swelling of sublingual soft tissues [14].

In our current case report, the child presented with feeding difficulties, leading to the insertion of an NG tube. However, just 3 days after the surgery, the patient was able to resume feeding normally. Ultrasound is the first imaging modality to diagnose sublingual epidermoid cyst due to its reliability and cost-effectiveness. The second level of imaging method is magnetic resonance imaging (MRI), specially used to find the exact location in relation to the geniohyoid and mylohyoid muscles of the cyst and plan surgery [15]. Differential diagnosis of sublingual epidermoid cysts includes ranula, thyroglossal duct cyst, lipoma, and infections of the floor of the mouth or salivary glands. The final diagnosis is usually confirmed with histopathology examination [16]. Surgical excision is the treatment of choice for epidermoid cysts in the oral cavity. The approach taken depends on the size of the cyst. For small sublingual cysts, an intraoral approach is typically selected, while for larger sublingual cysts, an extroral skin incision is preferred [17]. We chose an intraoral approach for this case, as the cyst was easily accessible through this technique and was successfully removed.

Author Contributions

Gelana Garoma Ararso: conceptualization, formal analysis, methodology, writing – original draft. **Demerew Dejene Dana:** data curation, validation, writing – review and editing. **Matewos Amare Fikire:** conceptualization, investigation, writing – original draft. **Mulugeta**

Temesgen Bonger: formal analysis, resources, validation. **Isa Salo Abdo:** formal analysis, resources, validation.

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The authors have nothing to report.

Ethics Statement

The ethical approval was not required for case reports at our Institution. Single case reports are exempt from ethical approval.

Consent

Written informed consent was obtained from the child's parent for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

All data supporting the findings of this case report are included in the manuscript.

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