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Case Report

Congenital Epulis in a 24-hour-old female neonate: A Case Report

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Abstract

Background

Congenital epulis is an extremely rare tumour of the oral cavity. If not treated appropriately with surgery, it can lead to mechanical obstruction, resulting in feeding difficulty and respiratory failure during neonatal life. Neonates with congenital epulis usually presents with mass protruding from the mouth, difficulty of breast sucking and difficulty of breathing. Diagnosis is usually suspected clinically and can be confirmed by histopathological examination of tissue excised from the mass. Multidisciplinary team management with surgical removal of the tumour is the main treatment option, and the outcome is usually excellent.

Case Presentation

We present a very rare case of congenital epulis in a 24-hour-old female neonate after presenting with a big protruding mass from the oral cavity, resulting in feeding difficulty. The histopathological examination revealed an upper alveolar ridge granular cell tumour, and the tumour was removed by surgery. After a few hours of surgery, the neonate was able to feed without any difficulty and was discharged in good condition after 3 days of observation.

Conclusion

Even though a begnin tumour, congenital epulis can lead to feeding difficulty by obstructing the oral cavity. With the appropriate multidisciplinary management, it can be corrected with a very good outcome.

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INTRODUCTION

Congenital epulis is an extremely rare tumour of the oral cavity with the potential of obstructing the oral cavity and airways, leading to feeding difficulty and respiratory failure, respectively (1, 2). This type of lesion had never been seen, and no report of congenital epulis had been made in this hospital before this neonate. It usually arises from the mucosa of the alveolar ridges of the jaws and occurs three times as often in the maxilla as in the mandible (3).

The histopathogenesis of congenital epulis has remained unknown in spite of a vast number of immunohistochemical and ultrastructural studies. Several theories have been proposed, including origins from odontogenic epithelium, fibroblasts, histiocytes, smooth muscle, nerve-related cells, endothelial cells, pericytes, myofibroblasts, and undifferentiated mesenchymal cells(4).

Clinically, it usually presents as a pedunculated nodule of less than 2 cm in size, although larger lesions have been reported, some even large enough to protrude from the mouth (3, 5). Most lesions are single, but multiple lesions may also occur on the same or different alveolar ridges (6). The management outcome with a multidisciplinary team of pediatrician, neonatologist, maxillofacial surgeon and anesthesiologist is usually excellent. In our hospital, the rarity of the case and the very good management outcome of the case drew attention and led us to report the case.

This study aims to expand the present knowledge on the prevalence of congenital epulis with its life-threatening complications by reviewing different literature and its outcome after treatment with a multidisciplinary team.

Case Presentation

This is a 24-hour-old female neonate born to a 20-year-old para II mother after amenorrhea of 9 months. The mother was having a regular antenatal care (ANC) follow-up at the local health center. There were no abnormalities detected. She was supplied iron sulphate and vaccinated twice. She had no danger signs of pregnancy. The delivery was uneventful and she gave birth to a female neonate weighing 3400 grams; the Appearance, Pulse rate, Grimace, Activity and Respiratory rate (APGAR) score was 8 & 9 at the 1^{st} and 5^{th} minutes, respectively. The neonate cried immediately after delivery.

The neonate was referred from the local Primary Hospital to Wollega University Comprehensive Specialised Hospital (WUCSH) with the complaint of a mass protruding from the oral cavity since birth. In addition to this, the neonate has difficulty of feeding because of the mass. Otherwise, the neonate has no cough, fast breathing, difficulty breathing, fever, or abnormal body movement. There is no bleeding or discharge from the swelling.

The mother has no previous history of delivery of a similar neonate. She has no history of alcohol drinking, cigarette smoking, or illicit drug use.

On physical examination: Well looking baby, Temperature=36.7C^{o}, Respiratory Rate=50 breaths per minute, Pulse Rate=110 beats per minute, Saturation of Oxygen=96%, Weight=3400 gram, Head Circumference=34 centimeter; A pink to brown, lobulated, pedunculated, firm, non-tender and smooth-surfaced mass measuring about 6*7cm on the anterior maxillary alveolar ridge. All adjacent tissues are normal on examination; The neonate

was alert, and neonatal reflexes (sucking, moro and routine) were normal; No other abnormal findings were present.

After we put the neonate on maintenance fluid made from 10% dextrose, she was investigated with complete blood count: white blood count=20,000 per cubic millimeter, neutrophil percentage =33%, lymphocyte percentage = 41%, hemoglobin = 14 gram/deciliter, platelet = 293,000 per cubic millimeter; Both abdominal ultrasound and echocardiography results were unremarkable.

With consultation of the maxillofacial surgeon, surgical excision was done under general anesthesia and an excision biopsy was sent to the pathology unit for possible histopathological examination. Histopathological examination shows a diffuse/poorly defined mass composed of sheets of cells; nests/ribbons separated by thin collagenous bands lined by stratified squamous epithelium, suggestive of upper alveolar ridge granular cell tumour.

The postoperative course was normal. Oral feeding was started after 6 hours of surgery, and she was feeding well. After 3 days of observation postoperatively, we discharged her when she fully started her mother's breastfeeding without any difficulty. The neonate was given an appointment at 2 weeks; after 2 weeks, she was fine and had no lesions in the oral cavity (Fig.1).

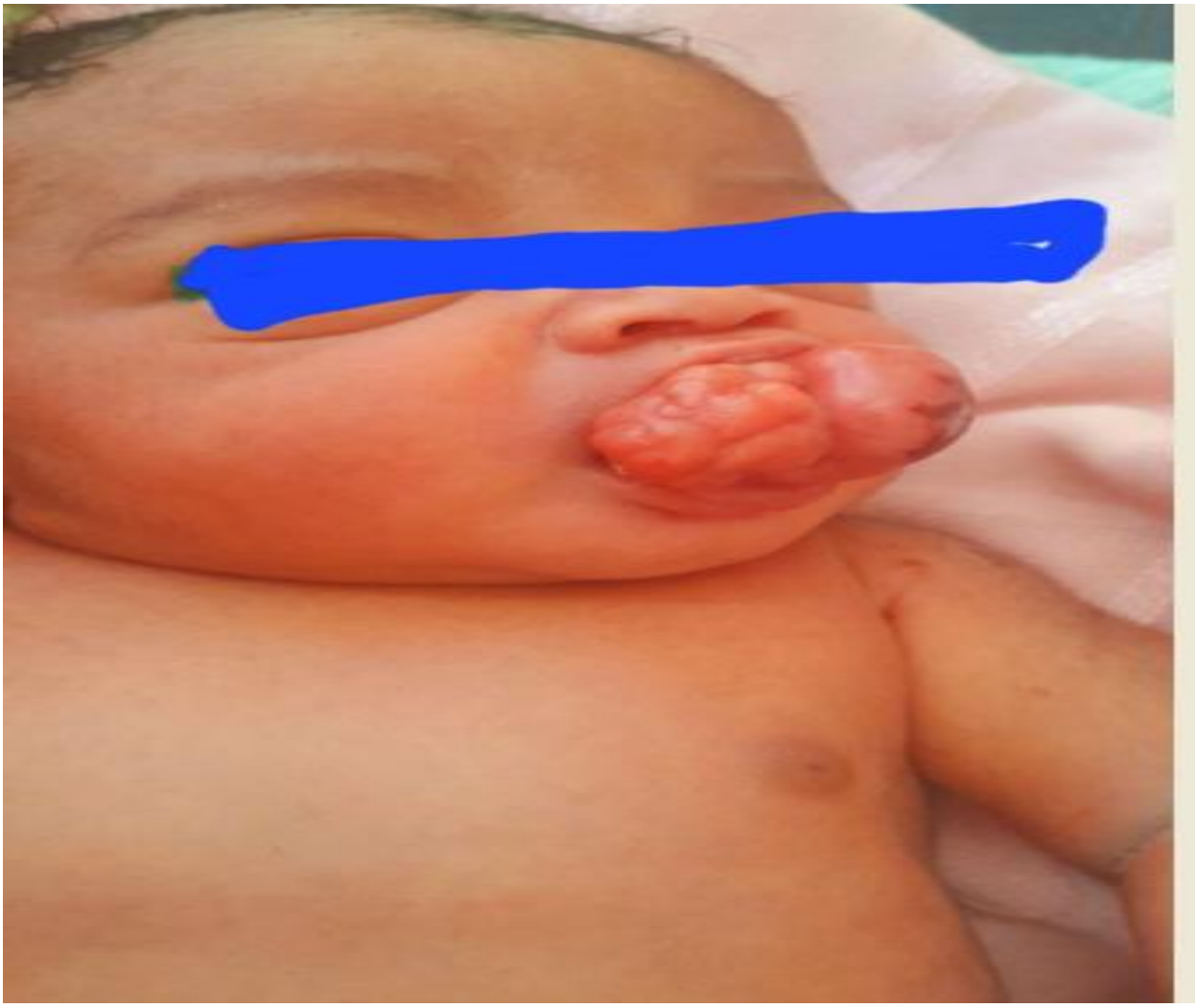

Figure 1. A neonate with Congenital Epulis before surgery.

DISCUSSION

Congenital epulis of the newborn is an extremely rare tumour of the newborn, also known as granular cell tumour or congenital gingival granular cell tumour because of its histologic features. This problem usually occurs in female newborns at a ratio of 8:1 (2). Our case is a 20-day-old female newborn. The exact etiology of congenital epulis is unknown, but several theories, such as origin from epithelial rests, undifferentiated mesenchymal cells, pericytes, fibroblasts, smooth muscle cells, nerve-related cells, and odontocytes, have been proposed. And also, the theory of endogenous hormonal stimulus has been proposed given female predominance (7).

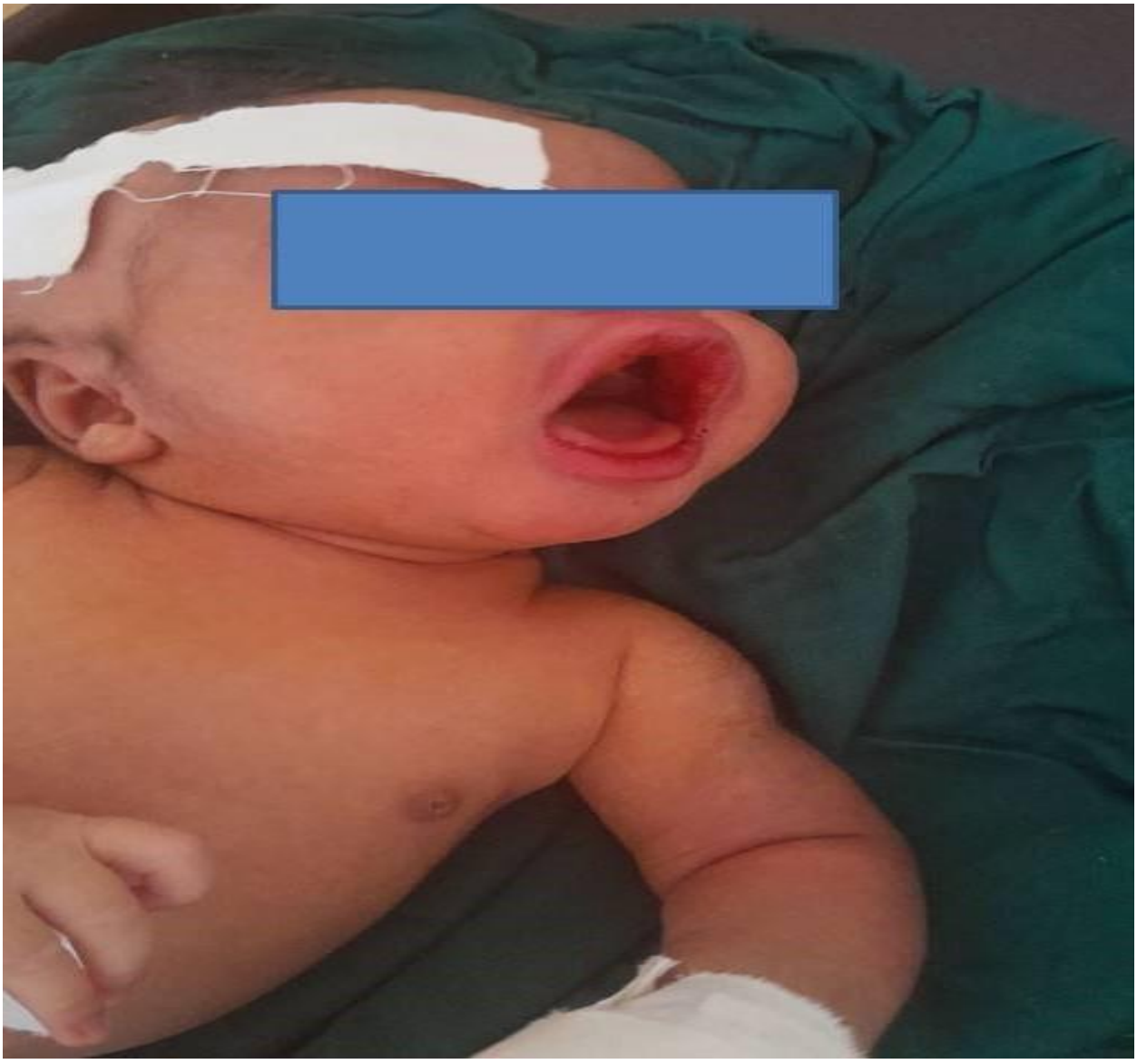

Figure 2. A neonate with Congenital Epulis after surgery

The diagnosis of congenital epulis is usually suspected clinically, after that can be confirmed with histopathological features of granular cell tumour. Hence, the interplay of neonatologists, dentists, otolaryngologists and pathologists are very important. The mass of congenital epulis is usually recognised immediately at birth or shortly after birth; the tumour presents in the alveolar mucosa as a smooth surface, singly/multiply, with red to pink in colour (7, 8). In our case, the mass was recognised at the age of 20 days; it was firm, pedunculated and pink from the maxillary ridge. The characteristic histologic features of congenital epulis are very important in distinguishing it from other masses like Epstein pearls, vascular malformations, and neuroectodermal tumours of infancy. These histologic findings include large round cells with granular, eosinophilic cytoplasm and small eccentric nuclei(9). The histopathologic finding in our case is a diffuse/poorly defined mass composed of sheets of cells, nests/ribbons separated by thin collagenous bands lined by stratified squamous epithelium

Surgical removal of the congenital epulis is usually mandatory, as spontaneous regression of the lesion is only rarely reported. This surgical removal will alleviate airway obstruction and feeding difficulties, and it will be easy to close the mouth(10, 11). In our case, surgical removal was done under general anesthesia with a maxillofacial surgeon.

Follow-up of the infant after surgery did not show any recurrence. The absence of recurrence even after incomplete excision, the possibility of spontaneous regression and the lack of a malignant counterpart all favour a non-neoplastic lesion(12).

CONCLUSION

Congenital epulis is a very rare tumour of the oral cavity with the common presenting feature of a mass protruding in the mouth. The mass usually results in difficulty feeding and difficulty breathing, with subsequent airway obstruction. Timely intervention of the tumour with surgical incision will completely relieve the problem of feeding and difficulty breathing.

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Authors' contributions

TS, HJ, ZF, GK, GH and DF were involved in the conception, study design, execution, acquisition of data, conducting the study, report writing, drafting, revising, or critically reviewing the manuscript; gave final approval of the version to be published.

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Declarations

Ethics approval and consent to participate

Wollega University Comprehensive Specialised Hospital has approved the publication of this case report.

Consent for publication

Written informed consent was obtained from the child's parent to publish this case report.

Competing interests

The authors declare that there are no competing interests.

References

1. Babu E, Kamalasanan G, Prathima G, Kavitha M. Congenital epulis of the newborn: a case report

and literature review. International Journal of Clinical Pediatric Dentistry. 2021;14(6):833.

2. Eghbalian F, Monsef A. Congenital epulis in the newborn, review of the literature and a case report. Journal of pediatric hematology/oncology. 2009;31(3):198-9.
3. Vered M, Dobriyan A, Buchner A. Congenital granular cell epulis presents an immunohistochemical profile that distinguishes it from the granular cell tumor of the adult. Virchows Archiv. 2009;454:303-10.
4. Lapid O, Shaco-Levy R, Krieger Y, Kachko L, Sagi A. Congenital epulis. Pediatrics. 2001;107(2):e22-e.
5. Aparna H, Jayanth B, Shashidara R, Jaishankar P. Congenital epulis in a newborn: a case report, immunoprofiling and review of literature. Ethiopian Journal of Health Sciences. 2014;24(4):359-62.
6. Uğraš S, Demírtaš Í, Bekerecioğlu M, Kutluhan A, Karakök M, Peker Ö. Immunohistochemical study on histogenesis of congenital epulis and review of the literature. Pathology international. 1997;47(9):627-32.
7. Jain N, Sinha P, Singh L. Large congenital epulis in a newborn: diagnosis and management. Ear, Nose & Throat Journal. 2020;99(7):NP79-NP81.
8. Merrett S, Crawford P. Congenital epulis of the newborn: a case report. International Journal of Paediatric Dentistry. 2003;13(2):127-9.
9. Küpers AM, Andriessen P, van Kempen MJ, van der Tol IG, Baart JA, Dumans AG, et al. Congenital epulis of the jaw: a series of five cases and review of literature. Pediatric surgery international. 2009;25:207-10.
10. Kayiran SM, Buyukunal C, Ince U, Gürakan B. Congenital epulis of the tongue: A case report and review of the literature. JRSM Short Reports. 2011;2(7):1-3.
11. Kumar RM, Bavle RM, Umashankar D, Sharma R. Congenital epulis of the newborn. Journal of Oral and Maxillofacial Pathology. 2015;19(3):407.
12. Kannan S, Rajesh R. Congenital epulis-congenital granular cell lesion: a case report. Journal of Indian society of pedodontics and preventive dentistry. 2006;24(2):104-6.