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Int J Case Rep Images 2023;14(2):32–36.
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# Congenital epulis in a female newborn

Mustafa Khawaja, Amer Hishmeh, Adel Asfour

## ABSTRACT

**Introduction:** Congenital epulis, also known as Neumann's tumor, is a benign soft-tissue lesion with an unclear cause. Congenital epulis manifests as a pink lump with a smooth surface in the mucosa of the maxillary or mandibular alveolar ridges. The lesion is more common in women and occurs more commonly in the maxillary alveolus than in the mandibular alveolus. Multiple theories have been proposed regarding the causes of congenital epulis; however, the etiology is still ambiguous.

**Case Report:** A 1-day-old female neonate was delivered via normal spontaneous vaginal delivery and referred to our hospital as an oral mass for surgical evaluation and intervention. It was discovered on a detailed prenatal ultrasound. Postnatal brain magnetic resonance imaging (MRI) was performed, the mass was characterized, laser excision was performed, and a histopathological examination confirmed the diagnosis. The patient underwent postoperative follow-up and was normal.

**Conclusion:** Congenital epulis is a rare, neonatal, congenital, and benign tumor. It requires removal by either surgical resection or laser excision owing to problems in feeding and respiration. Clinicians should always perform routine oral examinations for all newborns for the early diagnosis of this gingival tumor before it interferes with oral function.

**Keywords:** Benign tumor, Congenital epulis, Laser excision, Neumann's tumor, Oral mass

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Received: 08 July 2023
Accepted: 21 July 2023
Published: 08 August 2023

How to cite this article

Khawaja M, Hishmeh A, Asfour A. Congenital epulis in a female newborn. Int J Case Rep Images 2023;14(2):32–36.

Article ID: 101406Z01MK2023

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doi: 10.5348/101406Z01MK2023CR

## INTRODUCTION

Congenital granular epulis, also known as Neumann's tumor, congenital granular cell tumor, congenital granular cell myoblastoma, or congenital granular cell fibroblastoma [1], is a rare, benign soft tissue disease with an unclear cause. Newmann's tumor is the name given to the lesion after its discovery by Neumann, who published his findings in 1871 [2]. Lesions or tumors that protrude from the newborn's mouth are unusual. Although uncommon, these lesions should be removed immediately because of the risk of airway obstruction and death from asphyxiation [3]. A smooth, pink tumor known as a congenital epulis may appear in the mucosa of the maxillary or mandibular alveolar ridge [4]. This lesion occurs thrice more commonly in the maxillary alveolus than that in the mandibular alveolus and affects women 10 times more than men [5]. At a center in Wales, an incidence rate of 0.0006% was observed [6], and an Indian study found that epulis accounted for 10.8% of all oral lesions [7], with very few cases reported in the literature. In most cases of congenital epulis, only one lesion is present; however, in some cases, multiple lesions may occur. Although spontaneous regression has been documented in a few cases, these lesions are quite small, and the treatment of choice is surgical excision [8]. Multiple theories have been presented regarding the causes of congenital epulis; however, the etiology is still ambiguous. Herein, we present a case of a 1-day-old female neonate with congenital epulis of the right maxillary alveolar ridge.

## CASE REPORT

A 1-day-old female neonate delivered at (37+4 weeks) via normal spontaneous vaginal delivery at a local hospital, with a birth weight of 2.33 kg, was referred to the Neonatal Intensive Care Unit (NICU) at our hospital as a case of oral mass for surgical evaluation and intervention. The infant was delivered active, well, and crying, with an Apgar score of 8/9 and no maternal medical history of G2P1A1. The pregnancy course was uneventful. Detailed ultrasound (DUSS) was performed at 23 weeks gestational age (GA), and was repeated with free results at approximately 35 weeks GA. Routine US during the follow-up visit showed protruded macroglossia, and the family was advised to repeat DUSS. Repeated DUSS was performed and showed a large protruding mass from the oropharyngeal area (2.8 × 2.8 cm), consistent with oropharyngeal teratoma/epicanthus, or glossomegaly and needed evaluation after delivery. On physical examination after admission, a prorupted, solid, and non-cystic mass (3 × 4 cm) was observed attached to the right upper gingiva (Figure 1), while the remainder of the examination was uneventful. Brain MRI was performed on day 2 of life, and showed approximately (3.5 × 2.5 × 2 cm) heterogeneously enhancing soft tissue mass lesion protruding out of the oral cavity that appeared pedunculated originating anteriorly from the right upper roof. It returned a low T1 signal and heterogeneous low and high T2WI. It presented no calcification or pure lipomatous tissue and no extension into the nasal cavity, which appeared in the patient. The hard palate was intact. The lesion pushed the tongue to the left and slightly posteriorly, with apparent narrowing at the level of the oropharynx. Epulis, or congenital oral granular cell myoblastoma of the newborn, was primarily suspected. Other differential diagnoses included epicanthus and oral hamartomas. On day 3 of life, laser excision of the right gingival mass from the upper jaw was performed under general anesthesia without any complications, and the lesion was completely excised and sent for histopathological examination, which described the lesion as a single oval mass measuring 3.5 × 2.0 × 2 cm, revealing a polypoid tumor lined by atrophic squamous epithelium and composed of polyhedral cells with abundant granular eosinophilic cytoplasm and monomorphic vesicular nuclei with inconspicuous nucleoli (Figure 2). The tumor cells were negative for desmin, myogenin, S100, and SOX10 (Figure 3). No evidence of malignancy was found. On postoperative day 5, the patient was extubated and

Figure 1: Prorupted mass, attached to right upper gingiva, solid, non-cystic.

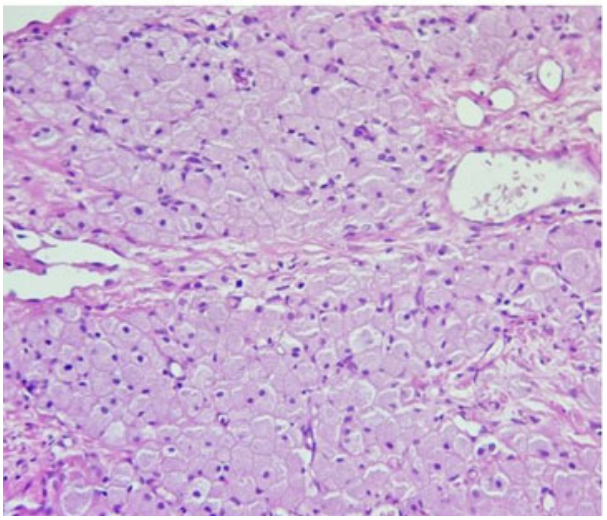

Figure 2: Hematoxylin and eosin (H&E) reveals a polypoid tumor lined by atrophic squamous epithelium and composed of polyhedral cells having abundant granular eosinophilic cytoplasm with monomorphic vesicular nuclei and inconspicuous nucleoli.

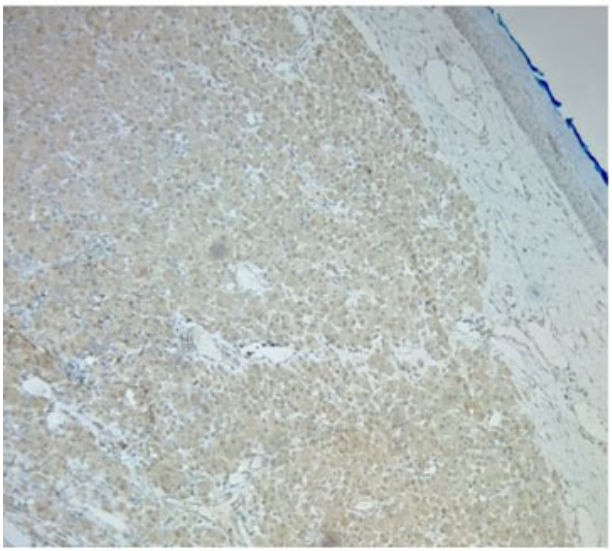

Figure 3: Immunostain; the tumor cells are negative for desmin, myogenin, S100, and SOX10 immunostains.

discharged on postoperative day 6. The patient could close the mouth, suck, and normal breast feeding was resumed. At 1-month postoperative follow-up, no signs of recurrence were observed and the parents reported an uneventful postoperative period.

## DISCUSSION

In this report, we describe a case of congenital epulis, a benign tumor that rarely occurs in infants. There are several names for this tumor, including congenital granular epulis, congenital granular cell myoblastoma, and Neumann tumor. The exact cause is unknown; however, hormones have often been implicated [1]. Studies using histology and electron microscopy have pointed to gingival stromal cells, including fibroblasts and histiocytes, as the source of lesions, suggesting a reactive theory of origin. This notion is supported by reports of spontaneous relapse and a lack of recurrence after incomplete resection [9]. The gingiva is a known source of congenital epulis [10]. Studies have demonstrated that the maxilla is affected more than the mandible, with a ratio of 3:1, and the tumor occurred eight times more frequently in women than in men [11, 12], which applies to the case of a newborn female with congenital epulis originating from the right maxillary alveolar ridge. Except in cases where the size is quite small and there are no symptoms, congenital epulis is typically diagnosed at birth or shortly after delivery. As no reliable signs of this disease are present before birth and the tumor rarely appears before the 22nd week of pregnancy, prenatal diagnosis is still challenging [1]. Through the use of ultrasonography techniques, the lesion can be diagnosed as early as the 36th week of pregnancy, enabling the delivery to be planned and surgical treatment to be performed as soon as possible afterward [13]. In our reported case, at GA 35-week, detailed ultrasound showed a large protruding mass from the oropharyngeal area, consistent with oropharyngeal teratoma/epicanthus, or glossomegaly, and needed evaluation after delivery. In most cases, there is a single hard tumor with a smooth surface that is either pink or red and non-tender to touch or may have many lobes. It is more common for congenital gingival granular cell tumors to present as a single mass; however, numerous lesions or simultaneous occurrences at several places in the oral cavity, including the maxilla, mandible, and tongue, have been reported [13, 14]. Congenital epulis does not grow after birth and is present at birth. Although there have been sporadic reports of spontaneous regression and recommendations for adopting a non-surgical expectant approach in cases of congenital epulis when there is no obstruction to feeding or respiration, spontaneous regression cannot be predicted [15, 16]. If a congenital epulis is large or numerous enough, it should be surgically removed as soon as possible to allow normal jaw and tooth development and to prevent feeding issues, and obstruction of the airways. In our case, feeding was difficult; however, breathing was not obstructed and there was respiratory distress. When used together, prenatal ultrasound and fetal MRI can help limit the differential diagnosis of oral masses [17], such as in our case, where the prenatal ultrasound revealed the lesion at the 35th week of gestation and helped distinguish congenital epulis from other masses of the oral cavity. However, there have been reports of cases in which surgical removal was performed in the newborn's first few days with no serious consequences, and there is no indication that this procedure had any effect on bone growth or tooth eruption [1, 18]. Depending on the number, size, and location of the lesions, surgery may be performed under local or general anesthesia. As in our case, the authors have indicated a preference for general anesthesia combined with nasotracheal intubation when the tumor size is large or numerous locations are involved [9, 19]. In our case, we performed laser excision of the lesion under general anesthesia because of its large size. Histopathological examination confirmed that the lesion was consistent with congenital epulis. Histological examination of the tumor specimen revealed a polypoid tumor lined by atrophic squamous epithelium and composed of polyhedral cells with abundant granular eosinophilic cytoplasm and monomorphic vesicular nuclei with inconspicuous nucleoli. In addition, with immunostain it was S100 negative, in contrast to the adult granular cell tumor which is S100 positive.

## CONCLUSION

Congenital epulis is an uncommon benign congenital tumor in newborns. Despite the possibility of spontaneous regression, feeding and breathing problems necessitate removal either through surgical resection or laser excision. Owing to its rarity and clinicians' lack of awareness, the tumor has been misdiagnosed before surgery. Consequently, clinicians should always perform routine oral examinations in all infants to detect this gingival tumor early before it starts affecting oral function.

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## Author Contributions

Mustafa Khawaja – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Amer Hishmeh – Conception of the work, Design of the work, Analysis of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Adel Asfour – Design of the work, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

## Guarantor of Submission

The corresponding author is the guarantor of submission.

## Source of Support

None.

## Consent Statement

Written informed consent was obtained from the patient for publication of this article.

## Conflict of Interest

Authors declare no conflict of interest.

## Data Availability

All relevant data are within the paper and its Supporting Information files.

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**Article citation:** Khawaja M, Hishmeh A, Asfour A. Congenital epulis in a female newborn. Int J Case Rep Images 2023;14(2):32–36.

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