

Congenital Epulis: A Surprise in the Neonate

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SOMMAIRE

La vue d'un nouveau-né atteint d'épulis congénital peut être saisissante pour les parents et les professionnels de la santé impliqués dans les soins néonataux. Ces tumeurs qui se situent dans la bouche des nourrissons peuvent être extrêmement larges et occuper une grande partie de la cavité buccale, entraînant un risque d'obstruction des voies respiratoires ou des problèmes d'alimentation. Les dentistes devraient savoir reconnaître ces tuméfactions car ils peuvent être appelés à consulter et à fournir des informations aux parents et aux autres praticiens au sujet de ces lésions.

Mots clés MeSH : gingival neoplasms/complications; gingival neoplasms/congenital; granular cell tumor/congenital; infant, newborn

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The congenital granular cell tumour of the newborn, also known as congenital epulis, is rare. It occurs on the gingiva of the anterior alveolar ridge of the jaws. These lesions behave in a benign manner and no recurrent or metastatic lesions have been reported.¹ They are seen 3 times more frequently in the maxillary alveolus than in the mandibular alveolus and the female to male ratio is 10:1.¹⁻³ The typical location is the alveolar ridge of the maxilla near the canine, but the mandibular region can also be involved.³

The etiology of the condition is unknown. Several theories have been suggested, namely, myoblastic, odontogenic, neurogenic, fibroblastic, histiocytic and endocrinologic.^{1,2} There are usually no associated dental abnormalities or congenital malformations,⁴ except for occasional reports of a hypoplastic or absent tooth and the possibility of mild midface hypoplasia.^{4,5}

The tumour presents in the alveolar mucosa as a smooth-surfaced sessile or pedunculated mass with a normal to reddish colour.^{6,7} Its size varies from several millimetres to a few centimetres in diameter, and it may interfere with respiration or feeding.^{8,9}

The diagnosis is usually clinical, although difficulties may occur when the index of suspicion is low or when the origin of the tumour is hard to determine. In such cases, the differential diagnosis is wide and imaging has a contributing role to play. Antenatal ultrasonographic features of congenital epulis have been described sporadically,¹⁰⁻¹³ but postnatal ultrasonographic findings have seldom been described. Correlative prenatal ultrasound and postnatal magnetic resonance imaging (MRI) findings have been reported.⁴

Surgical excision is generally indicated and no recurrences have been reported. Spontaneous regression of the lesion is rare.¹⁴ Histologically, the lesion is similar to the granular cell tumour, although pseudoepitheliomatous hyperplasia does not occur in the congenital epulis. Thus, the tumour mass comprises sheets of large, closely packed cells showing fine, granular, eosinophilic cytoplasm.¹⁵

This report documents the presentation and management of a congenital granular cell tumour of the maxillary alveolar ridge found in a newborn and treated with prompt surgical excision.



Figure 1: Appearance of a large mass of the oral cavity arising from the gingiva of the anterior maxilla in a neonate female.

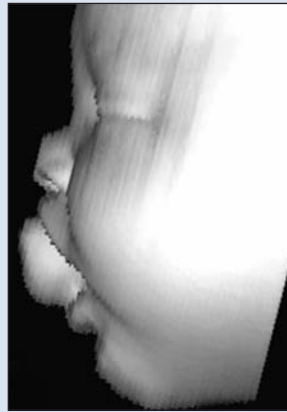


Figure 2: Three-dimensional CT scan showing the large mass protruding through the mouth.

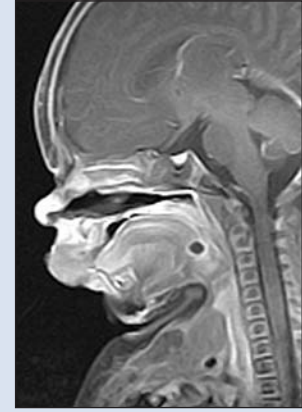


Figure 3: T1 weighted MRI image in the sagittal plane showing a mass attached to the anterior maxillary gingiva.



Figure 4: Lesion ready for removal in the operating room with patient's airway secured using an oral endotracheal tube.



Figure 5: Excised pedunculated lesion.

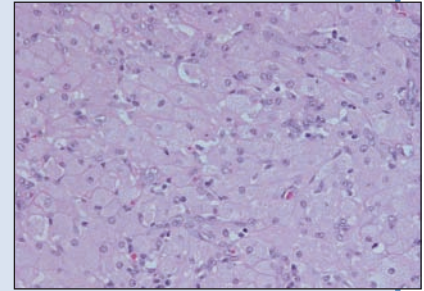


Figure 6: Granular cells present in the histologic specimen.

Case Report

A newborn girl was referred immediately after delivery for examination of a mass protruding from her mouth (**Fig. 1**). The pregnancy was normal and vaginal delivery occurred at 37 weeks. An ultrasound performed in the 29th week of gestation showed no abnormalities. No family history of hereditary diseases was reported.

On clinical examination, a midline, pedunculated, 3-cm-diameter round soft tissue mass exhibiting a smooth erythematous surface and located in the midline was found to be attached to the anterior gingiva of the maxilla. The mass prevented normal closure of the mouth and interfered with breastfeeding. The mass posed no immediate airway concerns. Feeding by a nasogastric tube was instituted.

General physical examination, including laboratory tests, were otherwise normal. Conventional ultrasonography with Doppler imaging showed a nonhomogeneous, solid, space-occupying lesion measuring 3 cm. The origin and extension of the mass could not be confidently determined based on ultrasonographic findings alone.

Computed tomography (CT) (**Fig. 2**) and MRI (**Fig. 3**) were performed to determine the extent and characteristics of the soft tissue mass. Both techniques revealed a lobular well-defined mass arising from the maxillary ridge, displacing the upper lip, without involvement of the unerupted teeth and without extension into the soft palate or intracranially. There was no significant enhancement of the lesion indicating that it was probably not vascular in nature.

Although congenital epuli can complicate general anesthesia by interfering with endotracheal intubation, this was not a factor in this case. The lesion was gently pushed to the side, the airway was visualized and an oral endotracheal tube was inserted (**Fig. 4**). The lesion was completely excised under general anesthesia, with minimal intraoperative hemorrhage (**Fig. 5**). Regular oral feeding was initiated immediately after surgery and was well tolerated. The infant was able to breastfeed on the third day after surgery and was discharged with her mother on the fifth day. At 2 weeks after surgery, the patient was reviewed and was noted to be thriving and gaining weight.

Histologic examination of the specimen revealed an unencapsulated lesion covered with squamous epithelium. The lesion was composed of homogeneous cells with granular eosinophilic cytoplasm and basophilic centrally located nuclei (Fig. 6). These findings were consistent with congenital granular cell tumour of the newborn.

Discussion

Congenital epulis, also known as granular cell tumour of the gingiva, congenital granular cell myoblastoma, or Newmann's tumour, following the first published case,¹⁶ is encountered exclusively in newborns. The tumour usually arises at the future site of the maxillary canine or the lateral incisors, but the unerupted teeth are not involved. The etiology remains unknown and controversial. Congenital epulis differs from other granular cell tumours encountered in adults by its exclusive origin from the neonatal gingiva, the scattered presence of odontogenic epithelium, the more elaborate vasculature and the lack of interstitial cells with angulate bodies.¹⁵

The clinical presentation consists of a lobular or ovoid, sessile or pedunculated swelling covered by a smooth mucosal surface, usually in the maxilla. A provisional diagnosis is often made clinically at birth and is confirmed histologically. Although the histogenesis of congenital epulis is not certain, it is thought to be a non-neoplastic, degenerative or reactive lesion.¹⁷⁻¹⁹ Its distinct visual presentation usually allows for direct clinical examination at birth. When the lesion is large and interferes with feeding and breathing, the treatment is simple surgical excision under either local or general anesthesia. Complete surgical excision is curative. Recurrence following incomplete excision has not been reported,⁹ making wide surgical excision unnecessary. Some very small lesions have reportedly undergone spontaneous regression.^{14,20}

Imaging in cases of congenital epulis may be important, especially for antenatal diagnosis using ultrasound¹⁰⁻¹³; the earliest reported case was identified in a 31-week-old fetus.¹⁴ In our case, ultrasound performed in the 29th week of gestation did not reveal any abnormality, suggesting that the tumour may have become apparent only at a later stage.

The CT and MRI findings of congenital epulis in our patient were in accordance with previous descriptions.¹⁴ Accurate mapping of the lesion with MRI ruled out maxillary and intracranial extension and was thus an important part of the preoperative planning.

The differential diagnosis of a large mass in the fetal or neonatal oral cavity should include such congenital malformations as encephalocele, dermoid cysts or teratoma and benign and malignant neoplasms including hemangioma, lymphatic malformations, melanotic or pigmented neuroectodermal tumours of infancy and rhabdomyosarcoma.

The congenital epulis is an oral mass that presents at birth in neonates. The lesion likely develops late in utero as it is often not detected on antenatal ultrasound. While the lesion is visually impressive and equally distressing, it is ultimately a benign lesion. If there is any fear of airway obstruction or difficulty with feeding, then prompt surgical treatment is necessary. Dentists may be consulted initially regarding such cases and should be aware of the potential for airway compromise and familiar with the differential diagnosis. ➤

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