Congenital Epulis: Unusual Etiology of Airway Obstruction and Feeding Failure in a Newborn

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Abstract

Title: Congenital Epulis: Unusual Etiology of Airway Obstruction and Feeding Failure in a Newborn
Objectives: Review congenital epulis; Its presentation and management.
Study Design: Case Report
Methods: Description of a newborn presenting with an obstructing oral mass. A review of the literature is included.
Results: Congenital epulis is a rare oral lesion that may result in airway obstruction and/or feeding failure bringing the mass to the attention of subspecialists.
Conclusion: A congenital epulis may present as a solitary alveolar mass in the newborn. Females are affected more often than males. Airway obstruction and feeding failure may evolve depending upon the size and location of the lesion. Physical examination, radiographic evaluation, and pathologic review are useful in its evaluation and diagnosis.
Histopathologically, special stains assist in the differentiation of the lesion from other solid tumors. Early intervention relieves airway obstruction and enables feeding success.

Introduction

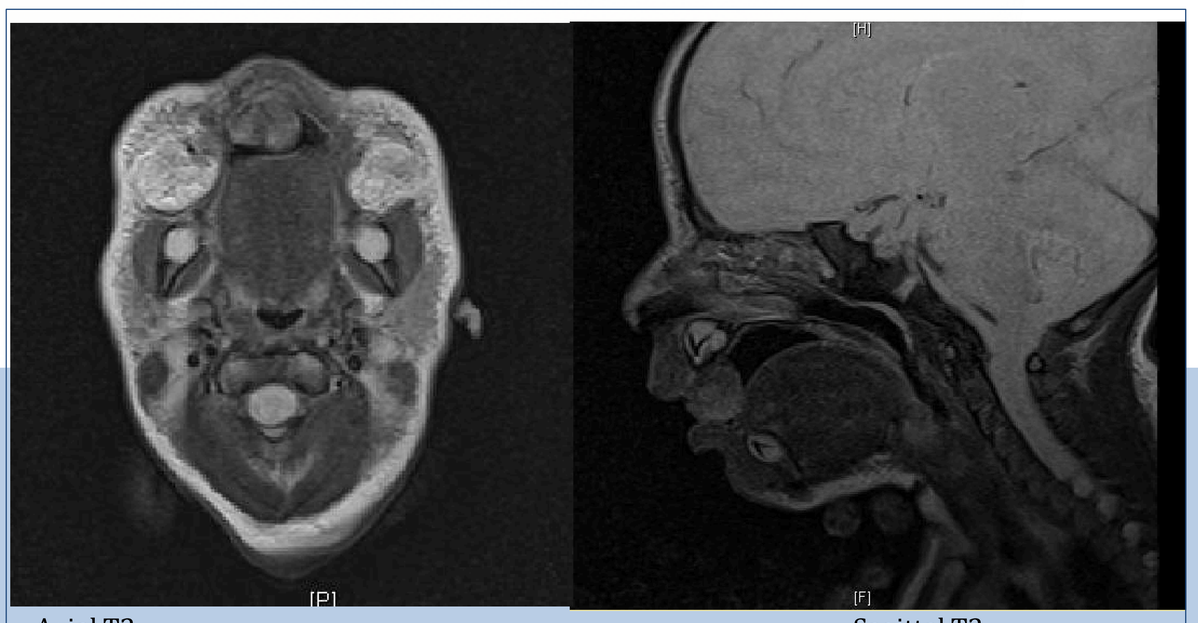
A congenital epulis is a rare lesion that may present in the newborn. Respiratory obstruction secondary to a congenital epulis may lead to asphyxia during the perinatal period. The lesion thus may present as a surgical emergency. Also known as a granular cell tumor, it may originate from either the maxillary or mandibular gingivae. The maxillary gingiva is more frequently involved. A female predilection has been noted. Few cases have been reported in the literature. We report a newborn with feeding challenges secondary to an identified congenital maxillary epulis. Early evaluation, diagnosis, and surgical intervention facilitated feeding success. Herein we present this case and provide a review of the literature.

Case Report

NM is a 2.8 kg female delivered at 40 weeks' gestation. On delivery, an oral mass was identified. Breastfeeding was initiated but was not successful due to the size of the lesion. Attempts at breastfeeding abraded the mass and led to local bleeding. The mass also excoriated the mother's nipple. On examination, the mass measured 2 cm x 2 cm and nearly obstructed all of the patient's mouth. The flesh colored, pendulous, mass was firmly adherent to the right maxillary margin (Figure 1).The palate was otherwise intact.

In evaluation, magnetic resonance imaging (MRI) of the face (Figure 2) was obtained and it revealed an exophytic 1.5 cm mass emanating from the right maxilla along the anterior inferior margin. T2 images of the mass were heterogeneous and T1 images were isodense. There were no dental elements within the lesion, but fibrofatty, cartilaginous, and mucosal elements of the alveolus were present. After discussion with the family, surgical excision was recommended due to the lesion's obstructive nature.

Description of Procedure: General endotracheal intubation secured the infant's airway for excision of the mass. Closer examination of the lesion evidenced a 2 x 2 cm lesion with a 1 cm base that contained two small veins and a small artery. Bovie electrocautery was used to incise the anterior mucosa. The incision was then extended laterally around the base thus freeing the posterior aspect of the lesion from the alveolar margin. The base was then lysed to the level of the 3 small vessels. Each were grasped and sequentially cauterized, thus releasing the lesion from the verge of the alveolus. The specimen was identified and sent for permanent pathology.



Axial T2 Sagittal T2

Figure 2: MRI Face wo contrast

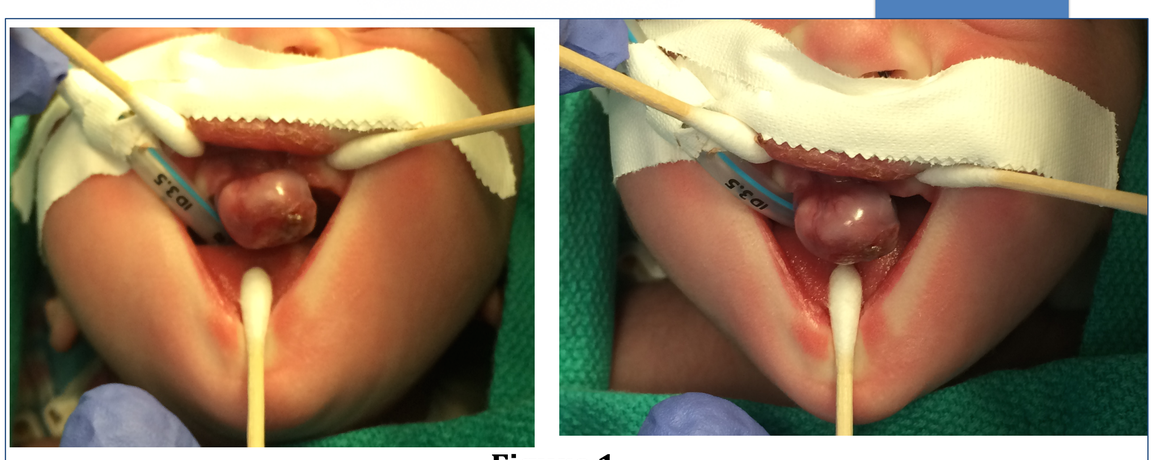
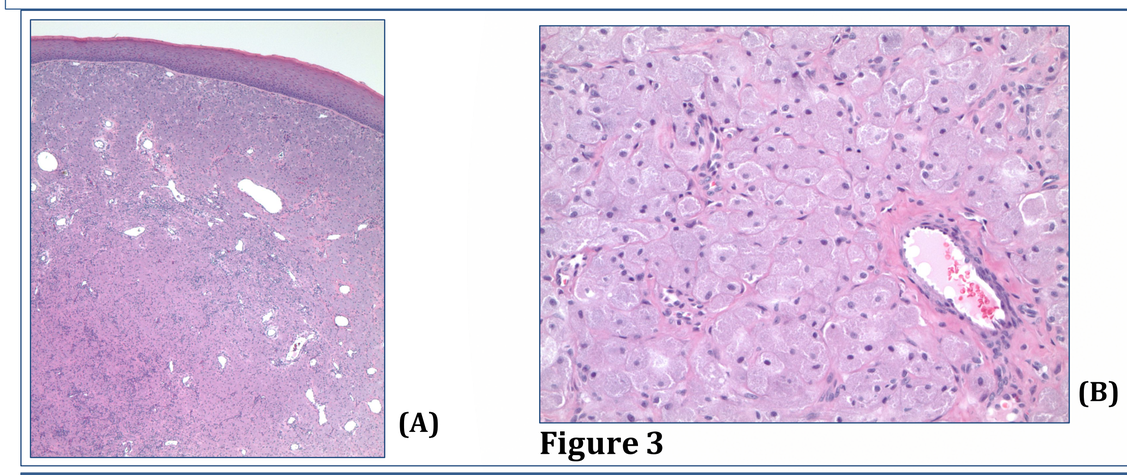


Figure 1

Results

The pathological specimen was a maxillary congenital granular cell tumor (congenital epulis).
Pathology slides (Figure 3):
(A) H&E stain, 4x: There is a subepithelial proliferation of cells with abundant eosinophilic cytoplasm. Note the absence of hyperplasia of the overlying squamous epithelium and the prominence of vascular structures.
(B) H&E stain, 20x: The tumor cells have abundant granular cytoplasm (low N/C ratio) and small uniform nuclei.
The patient was evaluated post-operatively and was tolerating breastfeeding without difficulty. No intraoperative or postoperative complications.



(A) (B)

Figure 3

Discussion

Congenital epulae or congenital granular cell tumors are rare. The differential diagnosis includes a teratoma, odontogenic cyst and dermatofibrosarcoma protuberans. Evaluation includes a thorough history and physical examination. Radiographic evaluation with a MRI assists to delineate the extent of the mass and should be obtained prior to surgery to aid in planning of the surgical approach. Interestingly, masses have been identified by fetal three-dimensional ultrasound late in pregnancy at 36th weeks gestation and assist in preparing for potential airway issues upon delivery.
Congenital epulae may originate from the mandible, maxilla or tongue. Though reported numbers are small, these lesions are typically benign and do not recur. The cell of origin is debated. Varied reports implicate fibroblasts, pericytes, epithelial cells, myofibroblasts, undifferentiated mesenchymal cells, and cells of neural origin. Lesion evolution is not well understood, but it does demonstrate cellularity that is benign, degenerative, and reactive. Congenital lesions do not stain positive for S-100 differentiating them from adult granular cell tumors that do stain positive for S-100 and are of Schwann cell origin.
Management of these lesions is usually surgical. Indeed, timely surgical excision is warranted for relief of airway obstruction and for oral feeding success. A multidisciplinary team that included neonatologists, pediatric anesthesiologists, and airway management surgeons (otolaryngologists, craniofacial plastic surgeons, and/or oral and maxillofacial surgeons) provides optimal care for these newborns.

Conclusion

Congenital epulis is a rare benign oral cavity lesion. Depending upon size and location, it may lead to airway obstruction and feeding failure. Physical examination, radiographic evaluation, and pathologic review are instrumental in the evaluation and diagnosis. Special stains differential the lesion from other solid tumors histopathologically. A multidisciplinary approach to these lesions that includes surgical excision results in a favorable prognosis for the newborn with relief of airway obstruction and oral feeding success.

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