Congenital epulis: from birth to childhood

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Congenital epulis, a rare benign neonatal tumor, is found more commonly in females and can be a cause of extreme anxiety in parents and health professionals. There is a potential of abnormal dentin development in surgically treated patients. These children should be followed by a pediatric dentist.

A watchful approach rather than surgery is justified in small tumors because of the risk of abnormal dentition at the site of surgery. These cases should be followed by dentists if surgical resection is done.

Key words: congenital epulis, congenital granular cell tumor, newborn.

Congenital epulis, histologically a granular cell tumor, is a rare neonatal tumor. First described by Neumann in the 19th century[7], it has only been noted in newborns and is a different entity from other granular cell tumors. It occurs eight times more commonly in females, the reasons for which remain unclear. It usually appears as a single pedunculated mass arising from the anterior maxillary alveolar ridge, and can cause problems with feeding and respiration due to the mass effect. Spontaneous regression has been reported occasionally, but most cases undergo early surgical resection. Recurrence and long-term dental outcome have not been reported. We report a newborn girl with a large epulis that was surgically resected but later caused problems with development of dentition, and we review the current literature.

Case Report

A newborn girl was born at term by elective cesarean section due to maternal pre-eclampsia. A large swelling arising from the middle of the upper gum margin was noted (Fig. 1). The baby did not require any resuscitation or develop respiratory symptoms. No other congenital anomalies were found.

She received nasogastric tube feeding and underwent surgical and oncological multi-disciplinary assessment. Magnetic resonance imaging (MRI) showed a mass of mixed signal intensity arising from the anterior hard palate with no evidence of intracranial extension (Fig. 2, T-2 weighted image).

A 3.5 cm diameter mass on a narrow peduncle was excised on day four under general anesthesia without complication (Fig. 3). The postoperative period was uneventful and breast-feeding was established over the next few days.

The histopathology showed a large polypoidal mass that was partially covered by squamous epithelium. The epithelium covered the sheets of isomorphic granular cells supported by a delicate fibrovascular network without any evidence of cellular atypia.

She later developed problems with primary dentition at the site of the surgery. Hypoplasia of the upper left lateral incisor was noticed at about 12 months of age, and later, hypoplasia of the left canine tooth. She was discharged from dental follow-up with the reassurance that her permanent teeth should be normal. She was referred back to dental services at two years of age with discoloration of the left lateral incisor tooth, indicating non-viability, and an associated buccal swelling thought initially to signal possible recurrence of the epulis. She underwent general anesthesia for extraction of the hypoplastic teeth along with removal of the canine on the right side as a balancing extraction. The soft tissue swelling was excised, which showed only inflamed granulation tissue histologically.

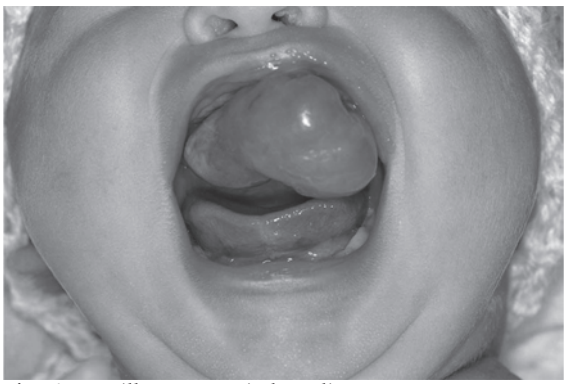

Fig. 1. Maxillary congenital epulis.

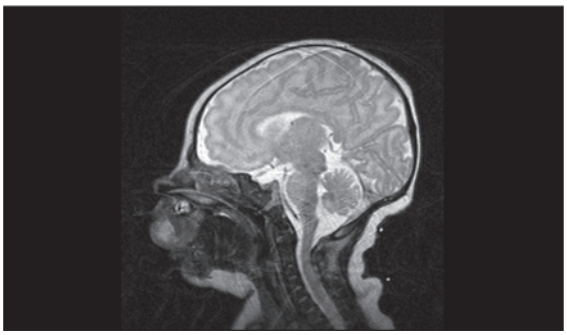

Fig. 2. MRI (T2-weighted image) of the same lesion.

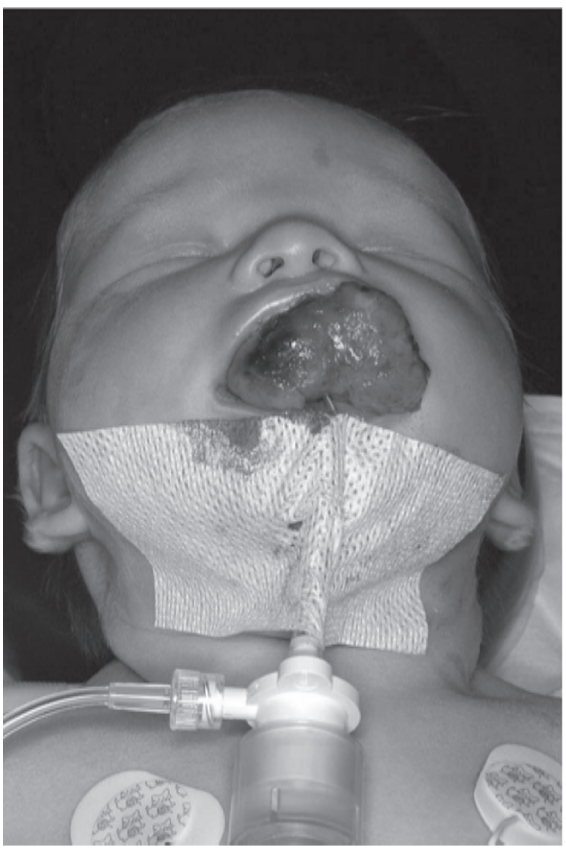

Fig. 3. Resection of the epulis under general anesthesia.

Discussion

Congenital epulis is a rare tumor of the newborn period, also known as granular cell tumor on the basis of histology. First described by Neumann in the 19th century[7], it usually arises as a single pedunculated mass form the anterior maxillary ridge. It is benign, but may cause significant parental anxiety as well as problems with feeding and respiration. In some cases, multiple tumors are noted[2], or the origin is mandibular[1], and it may be diagnosed on antenatal ultrasound[4]. It has a female predilection with an 8:1 ratio, but the biological basis for this has not been described. It is thought to originate from undifferentiated mesenchymal cells, fibroblasts, myofibroblasts, Schwann cells, or odontogenic epithelial cells, although the exact histogenesis is not known[3]. Spontaneous regression has been reported[5,8], but surgical excision is more commonly described, although large tumors may pose problems for endotracheal intubation[2,6].

We did not identify any reports of dental problems in the previous cases. This case developed dysplastic dentin and dentinal caries. Perinatal surgery poses a risk of damage to dental follicles developing in that region, so a watchful approach for spontaneous regression may be justified, especially when the swelling is small and does not interfere with feeding or breathing. Parents will benefit from appropriate counselling to explain the possible long-term effect on dentition, and a formal dental evaluation should be sought.

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