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Clinical Record

Congenital epulis

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Abstract
Congenital epulis or gingival granular cell tumour is a rare lesion occurring in the newborn, affecting females eight times more than males and arising more commonly from the maxillary than the mandibular alveolar ridge. We report the case of a five-day-old girl who presented to us with a large mass arising from the gingival mucosa of the mandible, causing feeding difficulty. Immediate surgical excision was followed by an uneventful recovery. The case is reported to make clinicians aware of this uncommon but easily treatable condition.

Key words: Gingival Diseases; Congenital

Introduction

Congenital epulis is a rare, benign tumour of the newborn. It is also known as congenital granular cell tumour or Newmann's tumour. This tumour arises from the gingival mucosa, either from the maxillary or alveolar ridges, and is typically seen as a mass protruding out of the newborn child's mouth causing interference with feeding and even respiration. The tumour is eight times more common in females than males and arises three times more often from the maxilla than the mandible.[1] The typical appearance and occurrence of the tumour on the alveolar ridge makes diagnosis easy. The usual presentation is of a pedunculated, nonulcerated, pink, firm mass ranging from a few millimetres to 9 cm in diameter. Simultaneous involvement of both maxillary and mandibular alveolar ridges has been reported in 10 per cent of cases.[2] The lesion runs a benign course and there have been no reports of recurrence or metastasis nor of any damage to future dentition. Cases of spontaneous regression have also been described.[3] The recommended treatment is immediate surgical excision. Because of its rarity and lack of awareness by clinicians the tumour has been misdiagnosed pre-operatively.

Case report

A five-day-old healthy girl was referred for a mass protruding from her mouth, which was causing difficulty in normal breast-feeding. The baby had been delivered normally at full term. There was no history of any drug intake by the mother during pregnancy or of any congenital abnormalities in the family. The birth weight was 2.8 kg. The child had no difficulty in respiration.

On examination, there was a pink, firm mass, measuring $5 \times 3 \times 2.5$ cm, arising from the lower alveolus and attached to the gingival margin in the region of the incisor teeth by a broad pedicle 0.5×2 cm in size (Figure 1). The surface showed superficial ulceration over a 1×1 cm area. A clinical diagnosis of congenital epulis was made.

The mass was excised under local anaesthesia. There was minimal blood loss. Histopathological examination (Figure 2) revealed sheets and clusters of cells with abundant eosinophilic, granular cytoplasm and monomorphic small, round-to-oval nuclei in a fibrous stroma covered by well oriented stratified squamous epithelium and showing a delicate plexiform capillary network. The tumour cells stained positive with Periodic-acid Schiff stain and were diastase-resistant.

The post-operative course was uneventful. The child was allowed to breast-feed on the day of surgery and was discharged the next day. At two-year follow up, there was no evidence of any recurrence, all the teeth had erupted normally and the child was in good health.

Discussion

The rarity of congenital epulis can be gauged by the fact that in the surgical pathology laboratory at the Medical College of Virginia, USA, only two such cases were seen over a period of 21 years.[4] In a review of the literature, Zuker and Buenecha found only 167 cases reported before 1993.[2] Dash *et al.*,[5] while reporting a case seen by them, added 11 more patients reported between 1993–2002.

Histopathologically, the lesions are commonly called granular cell tumours (GCTs) because of the presence of abundant coarsely granular cytoplasm. The first GCT was described by Abrikosoff in 1923 as a myoblastic neoplasm.[1]

Two distinct types of GCT have been identified in humans. Although there are minor histological differences between them, they may be divided for practical purposes on the basis of their site and age of presentation. The congenital epulis or congenital granular cell tumour (CGCT) presents as a distinct submucosal mass along the crest of the alveolar ridge in the incisor region of newborns. The maxilla is almost twice as often involved than the mandible but both jaws may be involved simultaneously. The tumour

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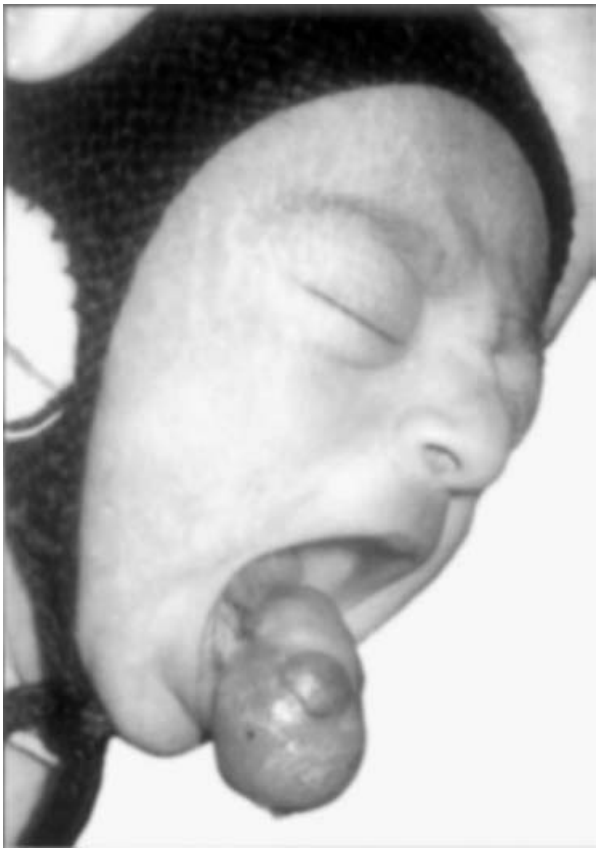

FIG. 1
The tumour.

is seen eight times more frequently in females than males. Simple excision is the treatment of choice. Recurrence is uncommon after surgical removal.[6] Lack of clinician awareness has lead to pre-operative misdiagnosis. The differential diagnosis includes haemangioma, lymphangioma, fibroma, rhabdomyoma and heterotropic gastrointestinal cyst.[7]

The second type of GCT is referred to simply as granular cell tumour or granular cell myoblastoma and is primarily a lesion of young adults, occurring more often in female and black patients. The most frequent sites of these tumours are the tongue and subcutaneous regions. These tumours have also been described in the soft palate, lips, floor of the mouth, larynx, trachea and bronchus.

According to Lapid *et al.*,[10] the CGCT contains cells which are virtually indistinguishable by light microscopy alone from those seen in the more common GCT of other sites. The distinguishing features, apart from the typical site and age of presentation, include histological characteristics such as plexiform arrangement of capillaries and the absence of pseudo-epitheliomatous hyperplasia of the overlying squamous epithelium. In addition, immunohistochemical studies have demonstrated the absence of S-100 protein, 75 KD nerve growth factor receptors, *trk* gene product and phosphotyrosine-positive cells in CGCTs and their presence in GCTs.[8]

While Schwann cells are considered to give rise to most GCTs, the histogenesis of CGCTs is still a matter of great controversy.[10] The various theories of its origin include myoblastic, neurogenic, odontogenic, fibroblastic and histocytic. However, Lack *et al.*[9] believed that the lesion is basically reactive in nature. In support of this is the fact that the tumour usually remains constant in size following birth of the affected infant, and on occasion the tumour even undergoes spontaneous regression. No local recurrence has been documented even when the tumour has been inadequately excised. The clinical behaviour of the tumour makes it clear that it is a benign lesion. Because of its female preponderance and spontaneous regression, it has also been postulated that the CGCT forms secondary to an intrauterine stimulus from fetal ovaries; however, Lapid *et al.*[10] found no evidence of oestrogens and progesterone receptors on immunohistochemical staining. Their findings on ultrastructural and immunohistochemical study support a mesenchymal origin. Dash *et al.*,[5] after

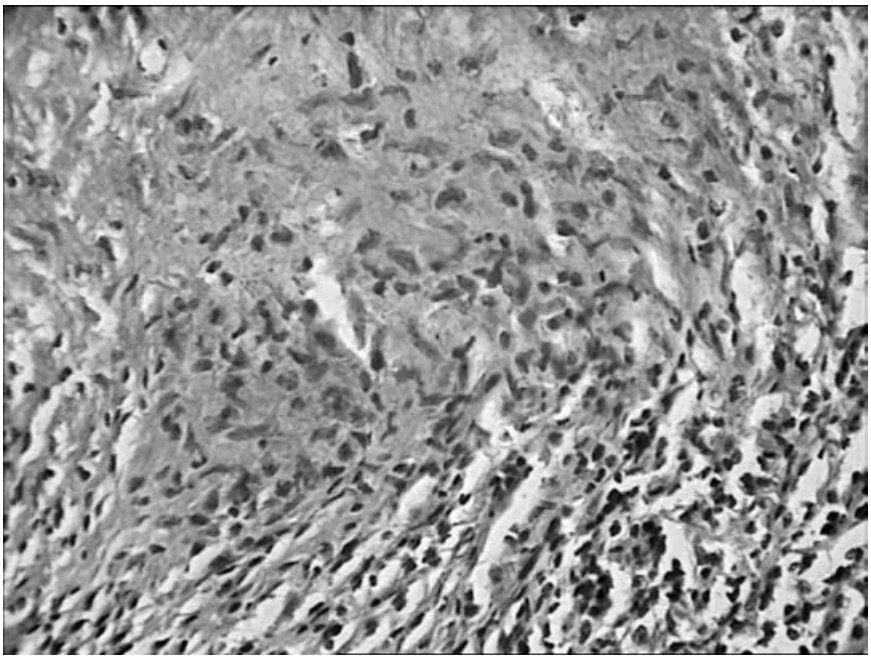

FIG. 2
Congenital granular cell tumour (H &E; ×40).

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reporting a case of congenital epulis, concluded that the biological behaviour of CGCT is compatible with embryonal hamartoma. They referred to it as 'congenital granular cell lesion' and not congenital granular cell tumour since it does not behave in a true neoplastic manner.

Once the clinician is aware of it, this benign lesion can easily be treated by conservative surgical excision.

Summary

Congenital epulis, or gingival granular cell tumour, is rare enough to justify report of its occurrence. It typically presents as a pedunculated, firm, pink mass a few millimetres to 9 cm in diameter on the alveolar ridge of newborns, more commonly on the maxilla than the mandible. It is eight times more common in females than males. Treatment is prompt surgical excision, and no recurrences are reported.

- **Congenital epulis or gingival granular cell tumour is a rare condition, typically presenting as a pedunculated, firm, pink mass on the alveolar ridge of newborns. The lesion is more commonly found on the maxilla than the mandible**
- **The condition is eight times more common in females than males**
- **Treatment is prompt surgical excision, with recurrence being rare**

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