Congenital gingival granular cell tumour

P J Anderson FDSRCS(Ed) FRCS P Kirkland FRCS
K Schafler FRCPath A L H Moss FRACS(Plast)

J R Soc Med 1996;**89**:53P–54P

SECTION OF PLASTIC SURGERY, 8 NOVEMBER 1994

Keywords: *congenital, gingival granular cell tumour, neonate*

Congenital gingival granular cell tumours are rare lesions which have only occasionally been reported in the UK. Clinical features are of a benign lesion which occurs almost exclusively in newborn, Caucasian females and the anterior maxilla is the commonest site. Treatment consists of local excision and is curative.

The terminology concerning this condition has been rather confused because of uncertainty regarding the histogenesis of these tumours and the similar histological appearance to adults granular cell myoblastoma occurring at other intraoral sites. The exact histogenesis of these tumours remains unresolved and they may be hamartomata.

We describe a new case occurring within the UK, which illustrates many of the common clinical features of the condition, with an accompanying literature review.

CASE HISTORY

A Caucasian female was referred a few hours after birth for urgent plastic surgical assessment with an intraoral swelling which was interfering with attempts at feeding. She had been born at term by a normal vaginal delivery following a normal pregnancy. No other abnormalities were found. It had been noted that she had a polypoid, congenital swelling $3 \times 3 \times 0.9$ cm which was superficially ulcerated arising from the anterior maxillary alveolus (Figure 1). The differential diagnosis included: odontogenic tumour, teratoma, neuroectodermal tumour and congenital granular cell tumour.

The initial management included intravenous fluids and nasogastric feeding. As oral feeding was clearly impossible for this child while this swelling remained, arrangements for early elective removal were made. The tumour was excised

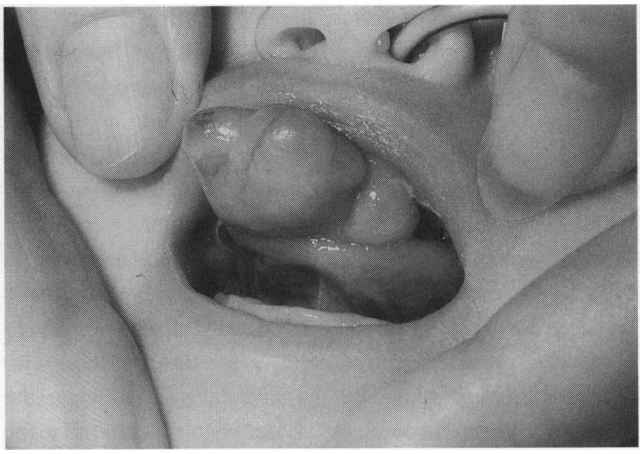

Figure 1 **The preoperative appearance of the tumour showing it arising from the anterior maxillary alveolus (note the nasogastric tube for feeding)**

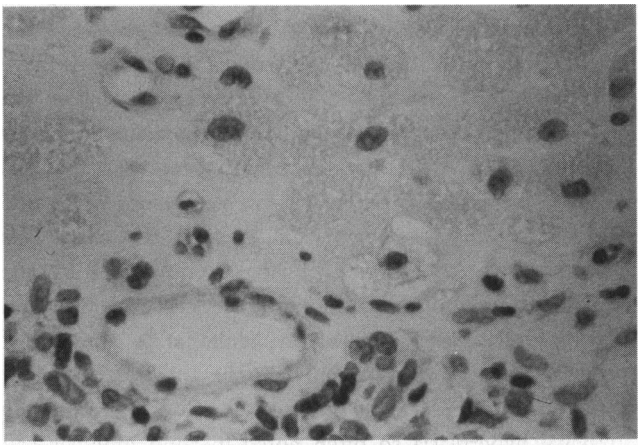

Figure 2 **Section through the tumour (×400) stained with haematoxylin and eosin, demonstrating characteristic large round cells containing granular cytoplasm and a central nucleus**

leaving the defect to heal by secondary intention under general anaesthesia, two days after birth. The patient was discharged home later the same day as surgery as oral feeding was readily established post-operatively.

Histopathology confirmed the diagnosis of a completely excised congenital gingival granular cell tumour (Figure 2). Immunohistochemical investigation confirmed the diagnosis.

Three months post-operatively the child is thriving with no evidence of recurrence.

DISCUSSION

Congenital gingival granular cell tumours have usually been sporadically presented as isolated case reports in British literature[1–4], since the first case was described in Germany in 1871[5]. The largest reported series comes from data collected throughout the USA over a period of 30 years, which reviews 21 of these lesions[6].

Departments of Plastic surgery and Pathology, Queen Mary's University Hospital, Roehampton SW15, England

Current address and correspondence to: Mr P Anderson, The Craniofacial Centre, The Hospital for Sick Children, Great Ormond Street, London WC1N 3JH, England

This case demonstrates several features which are characteristic of the condition. The child was female and Caucasian. The female to male ratio of this condition is 10:1[7], although a male has been reported in the UK population[2]. Where the racial origin of the child has been reported, only two cases worldwide occurred in non-Caucasians[6]. The mass occurred on the anterior maxillary alveolus, which has been reported as being affected twice as often as the mandible[6]. However, this lesion was unusually large at $3 \times 3 \times 0.9$ cm; they are usually less than 1.5 cm diameter[7]. Local excision is curative for the condition with no reported recurrences in the literature, even when the excision has been incomplete[6]. It has been reported that the natural history of the condition is for the lesion to spontaneously diminish in size[8], and that treatment should be conservative unless feeding was interfered with[9] (as in this case).

The terminology of this condition has been confusing due to continuing uncertainty regarding the histogenesis of the lesion. Both mesenchymal and odontogenic origins have been suggested[7], and the clinical behaviour has led to the suggestion that they may be considered hamartomata rather than neoplasms[9]. The term 'epulis' has been used, but this simply means swelling on the gingiva and lesions with quite different pathologies have been grouped together. This accurate diagnosis has been further complicated by the similar histological appearance to the oral granular cell 'myoblastoma' which occurs in adults at a number of intraoral sites, most commonly on the tongue. However, histological differences are noted in that the covering epithelial hyperplasia so often noted in adult granular cell tumour is absent in the congenital tumour[7].

Immunohistochemical staining can be of use in distinguishing the two conditions although there is some variation in the results obtained. In the noncongenital granular cell tumours there are usually positive responses to antigens for S100, and variable responses to myoglobin, myosin, actin, desmin, α-1-antitrypsin and muramidase. Congenital gingival granular cell tumour may stain positive for actin and myosin but not S100 as in our case, but other authors have found them negative for all markers[10].

In summary a case of congenital gingival granular cell tumour is reported which on reviewing the literature, exhibits many of the described clinical features but in addition is an unusually large example of this rare lesion.

REFERENCES

1 Middleton DS, Harvey WF. Congenital epulis. *Edinb Med J* 1933;**40**:257–65

2 Das PC. Congenital epulis. *Lancet* 1933; **225**:182–3

3 Hankey GT. Congenital epulis in a ten-weeks premature infant. *Proc R Soc Med* 1955;**48**:1015–17

4 Welbury RR. Congenital epulis of the newborn. *Br J Oral Surg* 1980;**18**:238–40

5 Naumann E. Ein Fall von congenitaler Epulis. *Arch Heilk* 1871;**12**:189

6 Lack EE, Crawford BE, Worsham GF, Vawter GF, Callihan MD. Gingival granular cell tumours of new newborn (congenital "epulis"). *Am J Surg Path* 1981;**5**:37–45

7 Lucas RB. *Pathology of Tumours of the Oral Tissues, 3rd edn.* London: Churchill Livingstone, 1976:229–34

8 O'Brien FV and Pielou WD. Congenital epulis: its natural history. *Arch Dis Child* 1971;**46**:559–60

9 Mann TP. *A Colour Atlas of Paediatric Facial Diagnosis.* 1st edn. Dordrecht: Kluwer Academic, 1989:18

10 Stewart CM, Watson RE, Eversole LR, Fischlshweiger W, Leider AS. Oral granular cell tumours: A clinicopathologic and immunocytochemical study. *Oral Surg Oral Med Oral Pathol* 1988;**65**:427–34

(Accepted 21 March 1995)