

## Congenital epulis: A rare benign baby lump

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### Abstract

Congenital epulis is a rare congenital tumor affecting the oral mucosa. It is usually seen in females and has no known syndromic association. It may cause the interference to feeding or airway obstruction, thus presenting as an emergency. Visually, it causes a lot of undue stress and anxiety to the parents and relatives. Multiple epulis occurs only in 10% of cases. Recurrence of the tumor, malignant change, or damage to dentition is not seen post-excision. Clinicians, particularly those working in the tertiary health care facilities, should be aware of this benign and easily treatable tumor of the neonate.

**Key words:** *Epulis, Gingival tumor, Granular cell tumor, Neumann's tumor*

**E**pulis or congenital gingival granular cell tumor was first described by Neumann in 1871 [1,2]. Epulis is a Greek term meaning “on the gum.” This tumor arises from the mucosa of the gingiva, most commonly from the anterior part of the maxillary alveolar ridge away from the midline [3]. Epulis is distinguished from other oral soft tissue tumors, by its maxillary or mandibular alveolar location (3:1). It is predominantly seen in females (8:1) and is usually solitary in nature. The recommended treatment is prompt surgical resection. Despite its aggressive appearance, malignant change, or damage to future dentition or recurrences of the tumor have not been reported [3]. Multiple lesions are rare occurring in only 10% of the epulis cases [1]. Around the 200 cases of epuli have been reported globally [4].

### CASE REPORT

A 3.1 kg girl baby was born at 39 weeks of gestation by normal delivery to a primigravida mother. Baby was referred to our institute with a growth in the oral cavity causing difficulty in feeding. The pregnancy was uneventful with normal antenatal scans in the first and second trimesters. There was no significant family history. The clinical examination revealed a full term female neonate with a 5 cm × 4 cm pedunculated growth attached to the anterior alveolar ridge of the mandible (Figure 1). The swelling was non-tender and firm in consistency. The mass prevented breastfeeding and closure of baby's mouth. The baby's airway was patent. The vitals of the baby were stable, and systemic examination was normal. Her routine laboratory investigations were non-contributory. Initial management included intravenous fluids and expressed

breast milk through orogastric tube, as direct breast feeding was not possible.

The tumor was excised under general anesthesia on postnatal day 2. Postoperative period was uneventful. Baby was started on direct breast feeds next day and discharged on day 5 of life (Figure 2). The gross specimen appeared pink, smooth surfaced, and soft to firm in consistency. Histopathology confirmed the diagnosis of congenital granular cell epulis. The baby was followed up at 45 days of life and was found to have gained weight and thriving well.

### DISCUSSION

Congenital epulis usually presents in neonate, although prenatal diagnosis with ultrasound has been reported in larger lesions as early as 26 weeks [5]. Congenital epulis may be single or multiple, sessile or pedunculated, usually pink and vary in size from a few millimetres to about 9 centimeters. The lesion is more commonly seen in the maxillary alveolar process, lateral to the midline in the incisor canine region. It is also seen in the mandibular region, and rarely on the tongue [1]. Prenatally a large epulis may cause maternal polyhydramnios due to impairment of fetal deglutition secondary to mechanical oral obstruction. Postnatally, it commonly interferes with airway and feeding. The actual incidence of epulis has so far not been estimated.

Histologically, the tumor is composed of nests of polygonal cells with abundant granular cytoplasm and small round nuclei. There is prominent vascular stroma and the lesion



**Figure 1: A 5 cm × 4 cm soft pedunculated erythematous non-mobile mass arising from the right side of the anterior mandibular alveolar margin in a 1-day-old female neonate**



**Figure 2: 3-day-old female neonate after removal of the mass**

is covered by continuous stratified squamous epithelium. Entrapped non neoplastic odontogenic epithelium may be seen in some cases.

The precise etiology of epulis is still uncertain. Intrauterine hormonal influence has been proposed to explain female preponderance and rapid growth during the third trimester. But this theory has not been proven so far due to absence of detectable oestrogen and progesterone receptors within the lesion [6,7]. There are several other theories suggesting its origin from undifferentiated mesenchymal cells, fibroblasts, myofibroblasts, histiocytes, Schwann cells or odontogenic epithelial cells. Immunohistochemical profiling has so far not been able to confirm the cell of origin. Vered et al. in 2009 suggested a local metabolic or reactive change as the cause for congenital epulis based on its lack of growth or spontaneous regression seen after delivery.

In our case, we considered a differential diagnosis of congenital epulis, leiomyomatous hamartoma, teratoma, granular cell tumor and haemangioma. Leiomyomatous hamartoma usually appear on the median anterior alveolar ridge or tip of tongue. Teratomas are seen on or under the tongue.

Granular cell tumors affect all age groups predominantly adults, usually donot affect the gingiva and show malignant change. Other tumors of the mandible like melanotic neuroectodermal tumor of infancy, embryonal rhabdomyosarcoma and lymphoma appear after one month of life and were hence ruled out. Other differentials that could be considered are rhabdoma, fibroma, granuloma, chondrogenic sarcoma and schwannoma [8]. These can be ruled out by histopathology.

Histologically of these differentials, congenital epulis closely resembles granular cell tumor. Both have large cells with eosinophilic granular cytoplasm. The difference is made on the basis of lack of pseudoepitheliomatous hyperplasia of the overlying squamous epithelium, plexiform arrangement of capillaries and positivity for vimentin and NKI/C3 in epulis. There is lack of S-100 protein, NGFR/p75 and inhibin alpha in immunohistochemistry in contrast to granular cell tumor [6,9].

Epuli are known to have spontaneous regression. Surgical excision is done if the lesion obstructs airway or interferes breastfeeding. Excision can be done under general or local anaesthesia. Local anaesthesia is preferred if the lesion is very small. Epuli were seen in children with polydactyly, goiter, maxillary hypoplasia, neurofibromatosis and polyhydramnios [8]. However so far no proven syndromic associations or dental abnormalities have been reported in congenital epulis [10].

## CONCLUSION

Congenital epulis is a rare, aggressive looking benign mass lesion of the oral mucosa, seen exclusively in neonates. Treatment is by simple excision in the immediate postnatal period. Congenital epulis does not have any known syndromic association or post-surgical sequelae.

Informed written parental consent has been obtained by using the patient's clinical details and photos for medical education and journal publication.

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