

Multiple Congenital Epulis: A Bombshell on the First Cry with Surgical Eviction by Erbium Laser

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Abstract

Congenital epulis also known as Neumann's tumor is a benign, rare congenital intraoral tumor that usually affects alveolar mucosa of neonates with a marked female predilection of 10:1 and site predilection for maxillary alveolus by 3:1. Very few cases have been reported in mandible and extremely rare < 10% cases have been reported with lesions in both the jaws simultaneously. Clinically tumor presents as a mass protruding through oral cavity of neonates with a potential to cause difficulty in feeding or respiration. Here we report an extremely rare case of multiple congenital epulis in a new-born in relation to maxillary and mandibular alveolus along with Epstein pearls and Bohn's nodules, which was managed successfully by surgical eviction of the mandibular lesion using Er Cr: YSGG and conservative management of smaller maxillary lesion, Epstein pearls, and Bohn's nodules. A review of literature on congenital epulis as well as erbium family of lasers should be considered as novel approach in management of paediatric oral surgical procedures.

Keywords: Congenital Epulis; Conventional; Laser; Mucocele; Surgery

Introduction

Granular Cell Tumor of Newborn also known as Neumann's tumor firstly reported by Neumann in 1871 usually affects the alveolar mucosa of neonates [1]. Surgical excision of the lesion is the recommended treatment. Although bohn's nodules and Epstein pearls are common entities but in literature their simultaneous occurrence with multiple congenital epulis in both the jaws has not been reported in scientific literature till date [2,21]. Here we report an extremely rare case of a new-born female patient, with multiple congenital epulis on maxillary and mandibular alveolus along with 2 Epstein pearls on midpalatine raphe and 2-3 Bohn's nodules on left maxillary alveolar gingiva [7,8].

A two days old female patient was referred to department of oral and maxillofacial surgery from paediatric NICU at Lifeline Hospital Panel, Mumbai. and then to Saisnehadeep Hospital, Koparkhaine Mumbai for evaluation and management of lesions in relation to her lower jaw that was found protruding from her mouth since the time of birth. Prenatal ultrasonography revealed no abnormality [9]. Family history for hereditary diseases was insignificant. Infant was delivered at full term 40th week of gestation without any difficulty. Birth weight was 2.571 kg.

A large lesion protruding from oral cavity was evident in relation to right side of mandibular alveolar ridge. The lesion didn't

cause any airway obstruction or respiratory distress hence there was no requirement of mechanical ventilation as the lesion was small in size however it prevented the normal closure of mouth and caused hindrance in breast feeding [7,8].

On extraoral examination there was no evidence of external facial asymmetry. Intraoral examination revealed two lesions, first on right maxillary alveolus and another on right mandibular alveolar ridge (Figure 1A). The lesion in relation to right maxillary alveolus was measuring 0.5×0.5 cm round, non-pedunculated, smooth, pink, non-pulsatile, and soft to firm in consistency. However, a feeding vessel was clearly evident over the surface of lesion (Figure 1C). Lesion in relation to right mandibular alveolus in canine- premolar region was measuring 3.5×2.5×2 cm soft tissue growth, pedunculated with 1 cm narrow and thin stalk attached to crest of alveolar ridge which was smooth, pink, lobulated, non-pulsatile, and soft to firm in consistency. Also 2-3 very small growths pearly white, 1-2 mm in diameter and firm in consistency over left maxillary alveolar crest on future primary molar site and 2 small, firm whitish growths, were evident in relation to mid palatine raphe at the junction of hard and soft palate that were suspected to be Bohn's nodules and Epstein's pearls, respectively based on their clinical presentation [11] (Figure 1B).

Figure 1: A. Mandibular congenital epulis. B. Maxillary congenital epulis. C. Epstein pearls on mid palatine raphe. D. Bohn's nodules. E. Notice the bilobularity and peduncle. F. Feeding vessel evident on surface of mandibular lesion.

On routine investigation her haemoglobin levels were 16.6 gm/dl and rest of parameters were well within normal limits. Vitamin K injection was given at birth and patient was started on Vitamin D₃ supplement. Standard immunization protocol was followed for the patient.

Consent form

Parents were informed about the entire procedure in advance and a written consent was signed.

Patient was operated under local anaesthesia at 15 days of her age using Er, Cr: YSGG laser (Waterlase iplus) at 0.5 watt, 20 pps, 1 air-water spray with gold hand piece, MZ6 tip and S mode which allowed smooth cut for excisional biopsy procedure (Figure 2A). A team comprised of Oral and Maxillofacial Surgeon, Pedodontist and Laser specialist performed the surgery with paediatrician and anaesthesiologist standby. A knee to knee position was acquired with elevated head end of the patient on assistant's lap. An extreme care was taken to avoid any chances of aspiration. There was negligible blood loss hence not sutured (Figure 2B). Excised lesion was stored in 10% formalin and sent for histopathological examination. Utmost care was taken to avoid aspiration by taking four major steps, patient positioning, high vacuum suction, decreasing the water flow in Laser and negligible bleeding because of Laser.

Oral feeds within 3 hours of surgery using a special dropper to avoid physical insult to the operated site. The patient was under observation and kept well hydrated for 24 hours in hospital. 2 doses of 2ml Syrup paracetamol was prescribed. Laser being bactericidal, no antibiotics were prescribed. The patient was discharged the next day and breast feeding resumed within 48 hours of surgery. Post-operative course and healing were uneventful as re-evaluated on 3rd and 7th post-operative day. Gingival re-epithelisation was completed within 3 days (Figure 2C). Patient evaluated on 1 month and 3 months' post-op for long term follow-up (Figure 3).

Figure 2: A. Surgical excision of lesion using Er Cr: YSGG laser. B. Surgical site immediately after excision. C. 1st post-operative day.

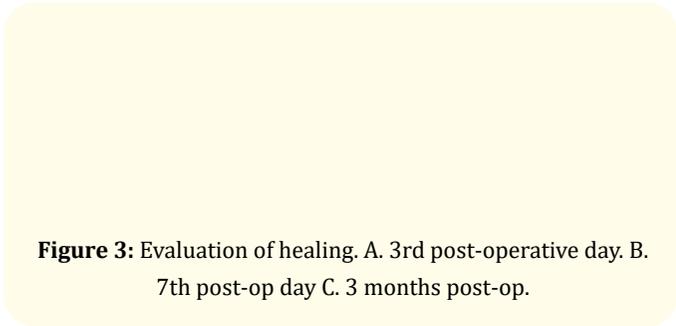


Figure 3: Evaluation of healing. A. 3rd post-operative day. B. 7th post-op day C. 3 months post-op.

Histopathological evaluation revealed it to be Congenital Granular Cell Tumor. Hematoxylin and Eosin (H&E)-stained sections of the lesion revealed the tissue lined by stratified squamous epithelium. Sub-epithelial layer was comprised of sheets and cords of polygonal cells having small round to oval nuclei with abundance of eosinophilic granular cytoplasm and plexiform network of capillaries. There was no evidence of spindling, necrosis, mitotic figures, nuclear pleomorphism, and granular dysplasia of malignancy [17,20]. (Figure 6 A to D). The smaller lesion in relation to right maxillary alveolus, Bohn’s nodules and Epstein pearls were managed conservatively as there was no hindrances and were totally asymptomatic [12-14]. Re-evaluation at 1-month post op revealed remarkable healing (Figure 4 and 5) and the parents were satisfied and happy with the result as they always wanted the treatment alternative to conventional surgery and GA.

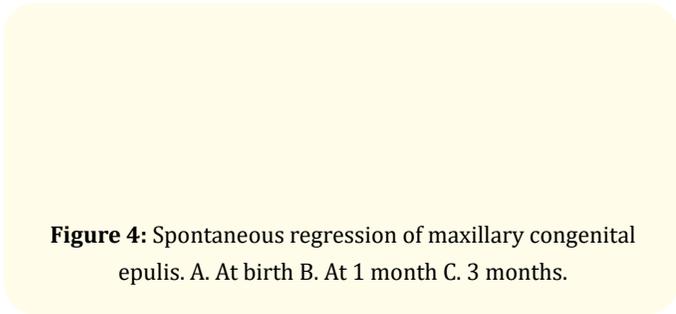


Figure 4: Spontaneous regression of maxillary congenital epulis. A. At birth B. At 1 month C. 3 months.

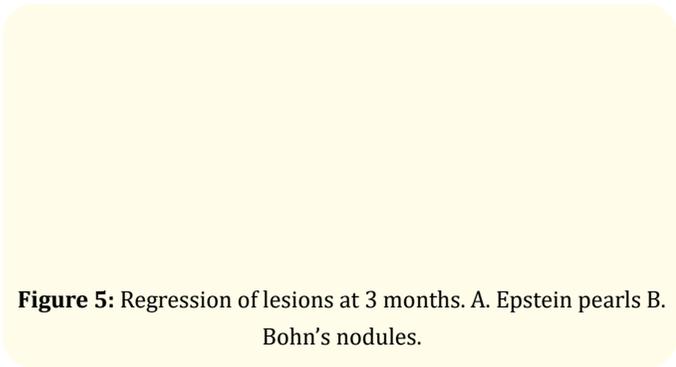


Figure 5: Regression of lesions at 3 months. A. Epstein pearls B. Bohn’s nodules.

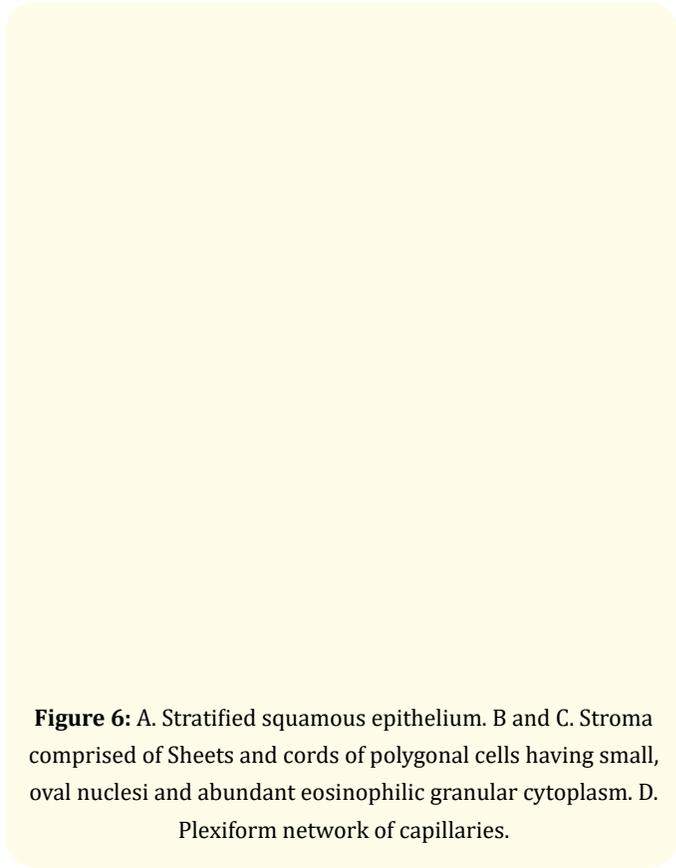


Figure 6: A. Stratified squamous epithelium. B and C. Stroma comprised of Sheets and cords of polygonal cells having small, oval nuclei and abundant eosinophilic granular cytoplasm. D. Plexiform network of capillaries.

Discussion

Congenital Epulis is a rare benign soft tissue tumor as per report by Zuker and Buenecha, who reported only 167 cases until 1993 arising from alveolar mucosa of maxilla or mandible [5]. Our case with its presentation as multiple congenital epulis with bohn’s nodules and Epstein pearls makes it the first case to be reported in literature.

Treatment of choice is surgical excision of lesion as soon as possible under general or LA using scalpel, electro cautery, and lasers. Local anesthesia can be considered for difficult intubation and for smaller lesions. In literature very few cases of spontaneous regression of the lesions has been reported in cases where there are no functional interferences. Regular monitoring of such lesions has been advocated as an acceptable clinical approach [18,19].

Lasers have revolutionized the entire medicine and surgical fraternity. AAPD advocates the judicious use of laser in providing management of soft tissue lesions in infants, children and adolescent [15,16]. CO₂ lasers were the first to be integrated into maxillofacial surgery for soft tissue surgeries. Primary effect of laser

on target tissue is photo thermal effect on water of target tissue. Since the soft tissues are mainly composed of water mainly through which water of tissues gets vaporized resulting into soft tissue ablation. Lasers are more precise and cause minimal thermal necrosis of adjacent tissues when compared with electrosurgical devices.

Erbium family - Er: YAG and Er Cr: YSGG laser with wavelengths 2940 and 2790nm, respectively are similarly effective in management of soft and hard tissues. They have shallow depth of penetration, high affinity for water, minimal reflective property. Early and most appropriate management strategy should be formulated with a well versed multidisciplinary team approach. Minimal thermal damage which make erbium family of lasers to be the ideal for paediatric procedures as compared to diode and CO₂ Laser [4,15,16]. Soft tissue procedures that usually require GA can be operated quickly, safely and with minimum or no local anaesthesia with very little water spray and gentle airflow that allows tissue cooling [16,20]. Major benefits of managing patients with erbium lasers are: 1. Bactericidal effect, thus resulting in sterilization of the area and thus eliminating the need of prophylactic or Postop antibiotics. 2. Immediate haemostasis 3. Reduced chair side time. 4. Suture less surgery. 5. Minimal postop discomfort. Thus erbium lasers have been considered ideal and superior to CO₂, Diode, and Nd: YAG lasers in management of paediatric oral surgical procedures.

Conclusion

To conclude proper examination of such rare lesions, appropriate diagnosis and formulation of a set of differential diagnosis should be done to avoid under/over treatment or any sort complications. Involving obstetric-gynaecologist, paediatrician, LASER Specialist, Pedodontist, oral and maxillofacial surgeon, radiologist and anaesthesiologist [9]. Erbium lasers enables a surgeon to perform surgical procedure on daycare basis. To best of our knowledge our case is the first case in literature which was treated by using Er, Cr: YSGG laser. Prime rationale of using laser in paediatric oral surgical procedures is to provide a bloodless, painless, and sutureless wound closure without general anaesthesia as a fearless substitute to scalpel or electrocautery to facilitate faster and efficient postoperative surgical wound healing hence considered as novel approach in management of paediatric oral surgical procedures.

Conflict of Interest

None.

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