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(S. Sembronio)

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Re: re: Condylectomy: treatment of recurrent unilateral dislocation of the temporomandibular joint in a patient with Ehlers-Danlos syndrome

Sir,

In response to the letter to the editor on our article entitled “Condylectomy: treatment of recurrent unilateral dislocation of the temporomandibular joint in a patient with Ehlers-Danlos syndrome”¹ we thank Sembronio et al² for their constructive comments. We apologise for not having referenced endoscopic techniques in our report, and we thank them for making us aware of their work.

In our opinion, disc pliation, either open or closed, would not have been appropriate in this case. The dislocation was a result of the anatomical relation of the condyle to the deeply set fossa rather than the disc. The disc pliation or eminectomy techniques described in the letter, therefore, may have not been appropriate in this case.

Our patient has been free of symptoms, and has regained full function and occlusion. MRI imaging is not necessary therefore for follow up, as all symptoms have fully resolved. Regarding occlusion, as a result of our protocol of intermaxillary fixation, the senior author has not experienced malocclusion in 11 cases of high condylectomy.

In summary, current established practice is based on series of case studies, some of which are decades old. We presented a case that had been managed differently to the set dogma of eminectomy, infracture of the zygomatic arch, and repositioning of the disc. Our technique resolved the disabling dislocation with full restoration of function and no complications.

Ethics statement/confirmation of patients’ permission

Not applicable.

Conflict of interest

We have no conflicts of interest.

References

1. Campbell SJ, Chegini S, Heliotis M. Condylectomy: treatment of recurrent unilateral dislocation of the temporomandibular joint in a patient with Ehlers-Danlos syndrome. *Br J Oral Maxillofac Surg* 2019;57:76–8.
2. Sembronio S, Tel A, Robiony M. Re: Condylectomy: treatment of recurrent unilateral dislocation of the temporomandibular joint in a patient with Ehlers-Danlos syndrome. *Br J Oral Maxillofac Surg* 2019 (In press).

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Congenital epulis: a rare case of feeding obstruction in a neonate

Sir,

Congenital epulis, also known as congenital granular cell tumour, is a rare, benign, neoplastic lesion of the newborn. It is often found on the anterior maxilla, has a strong female:male predilection (10:1) with a classic granular histopathological appearance.^{1,2} It is recognised as an uncommon cause of feeding obstruction in neonates.²

We present a case of a large, congenital epulis that obstructed feeding in a newborn child.

A 4-day-old baby girl was referred to the department with a lesion on the anterior maxilla that interfered with her feeding. She was born at term after an uncomplicated pregnancy, was otherwise fit and well, and thought to be feeding satisfactorily before she was discharged from hospital. The mother attended a scheduled appointment shortly after the birth, and expressed concerns about the baby’s ability to achieve an adequate oral seal while feeding.

On examination a large, pedunculated, firm, smooth, regular mass that measured 20 × 10 mm was seen at the anterior maxillary alveolus. A congenital epulis was suspected, so we infiltrated a small volume of local anaesthetic with adrenaline, applied a surgical vascular tie (Fig. 1), excised the lesion, and left the base to granulate.

Histopathological examination showed a lesion composed of sheets of large polygonal cells with eosinophilic granular cytoplasm, small central nuclei, and occasional odontogenic rests, and confirmed the diagnosis of congenital granular cell tumour (congenital epulis).

The baby was reviewed three weeks later. The excision site had completely healed and her mother commented on a pronounced improvement in feeding with an appropriate gain in weight (Fig. 2).



Fig. 1. Photograph of the surgical tie in situ.



Fig. 2. Photograph three weeks after removal of the tie (published with the permission of the patient's parents).

First described by Neumann in 1871 (quoted by Lack et al³), a congenital epulis is a rare but recognised lesion of newborn babies. It is likely to arise from cells of mesenchymal, neuroendocrine, or odontogenic origin, and the aetiology remains unknown. However, an endocrine theory has been proposed in view of the female prevalence.⁴

The differential diagnoses of congenital epulis include: leiomyomatous hamartoma, pyogenic granuloma, congenital haemangioma, and congenital fibrosarcoma. The lesion is not associated with any other congenital malformations.^{1,2}

The granular cell type identified in this case is the most common histological subtype, and granular cells were seen at the periphery of the lesion. No immunohistochemical analy-

sis was done, and none of the rarer features such as fibrosis, spindle-cell proliferation, or leiomyomatous change were seen.^{1,5}

Spontaneous regression can occur but was not an option in this instance because of concerns over feeding, and we favoured a narrow excision. Recurrence, even in the presence of involved peripheral margins (as seen in this case), is almost unheard of.⁵

Large congenital epulides are rare and can be a cause of considerable distress to unsuspecting parents. This case highlights the aesthetic and functional impact such lesions can have in the first few days of life, and the need for prompt diagnosis and minimal surgical intervention.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

Ethics approval not required. We obtained permission from the patient's parents.

References

1. Kujan O, Clark S, Sloan P. Leiomyomatous hamartoma presenting as a congenital epulis. *Br J Oral Maxillofac Surg* 2007;**45**:228–30.
2. Ben Hamouda H, Ayat A, Belaid L, et al. Obstructive congenital epulis. *Eur Ann Otorhinolaryngol Head Neck Dis* 2010;**127**:86–9.
3. Lack EE, Worsham GF, Callihan MD, et al. Gingival granula cell tumors of the newborn (congenital "epulis"): a clinical and pathologic study of 21 patients. *Am J Surg Pathol* 1981;**5**:37–46.
4. Bilen BT, Alaybeyoğlu N, Arslan A, et al. Obstructive congenital gingival granular cell tumour. *Int J Paediatr Otorhinolaryngol* 2004;**68**:1567–71.
5. Conrad R, Perez MC. Congenital granular cell epulis. *Arch Pathol Lab Med* 2014;**138**:128–31.

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Glossopharyngeal neuralgia: a case report

Sir,

Glossopharyngeal neuralgia is rare and affects the glossopharyngeal nerve and some regions that are supplied by the vagus nerve.¹ Patients report brief, intense episodes of paroxysmal pain in structures such as the soft palate, tonsillar fossa, posterior third of the tongue, oropharynx, auditory canal, middle ear, or angle of the mandible.^{1–4} Continuous neuropathic pain may also occur.² In 10% of cases the involvement of the vagus nerve results in parasympathetic symptoms such as bradycardia, hypotension, syncope, seizures, and asystole.^{2,3}

As a result of the numerous structures that may be affected, clinical presentations of glossopharyngeal neuralgia vary. Symptoms may be suggestive of many other conditions such as temporomandibular dysfunction,⁴ trigeminal

Box 1: Diagnostic criteria for glossopharyngeal neuralgia.

- A Repeated paroxysmal attacks of pain in the distribution of the glossopharyngeal nerve on one side
- B Pain has the following:
 - 1 Short duration (<2 minutes)
 - 2 Severe intensity
 - 3 Electric shock-like, shooting, stabbing or sharp in quality
 - 4 Precipitated by swallowing, coughing, talking or yawning
- C Not better accounted for by another diagnosis

neuralgia,⁴ superior laryngeal neuralgia,^{1,5} nervus intermedius neuralgia,⁵ and first bite syndrome. Occasionally, pain can even radiate to the eye, nose, chin, or shoulder,¹ which can further complicate the history. Patients may also struggle to localise or describe pain accurately, which affects the deep structures of the head and neck.⁵

A 78-year-old man was referred by his general dental practitioner with left-sided pain in the temporomandibular joint (TMJ). He complained of a two-year history of a constant dull ache in the region of the left ear and TMJ, which was interrupted by short episodes of intense, stabbing pain that occurred mainly while he was eating. Clinical examination showed no intraoral or extraoral abnormalities.

Updated diagnostic criteria for glossopharyngeal neuralgia have been published (Box 1), which include: repeated paroxysmal pain on one side, along the glossopharyngeal nerve; pain that is short-lived (under two minutes), severe, intense, sharp, stabbing and electric-shock-like; pain that is preceded by swallowing, yawning, talking, or coughing and; pain that cannot be explained by any other diagnosis.¹ Based on these criteria, we made the diagnosis. While it is mostly academic for medically-managed patients, accurate differentiation between trigeminal and glossopharyngeal neuralgia is still essential because of the variety of surgical options.

Many cases of glossopharyngeal neuralgia are idiopathic,² but imaging must be done to assess the potential causes such as vascular malformation, demyelinating diseases, intracranial and extracranial tumours, infections, injury, Chiari-I malformation, or Eagle syndrome.^{2,3,5} High-resolution magnetic resonance imaging is recommended for all patients,^{2,4} and computed tomography can be used adjunctively.⁵ Nasendoscopy is particularly useful to screen for local causes, such as malignancy in the oropharynx.³

The medical management of trigeminal and glossopharyngeal neuralgias are similar.² Current research supports the use of carbamazepine as a first-line treatment for all patients, unless contraindicated,^{2,4} and if it is poorly tolerated, oxcarbazepine can be used, as it has fewer side effects.^{2,4} Second-line treatment with baclofen or lamotrigine is supported by level C evidence,^{2,4} but there is also