

IN BRIEF

- Necrotizing sialometaplasia is an important lesion as it can be misdiagnosed as a malignancy.
- No treatment has been described previously either for the lesion or anaesthesia which may be associated.
- This is an unusual case since both bilateral occurrence and bilateral anaesthesia has not been previously reported.
- Since aetiopathogenesis is inflammatory, a trial of intralesional steroids was undertaken. Unfortunately this had no effect on the clinical course of the lesion, or the anaesthesia, which resolved spontaneously in 2 years.

Necrotizing sialometaplasia: An unusual bilateral presentation associated with antecedent anaesthesia and lack of response to intralesional steroids. Case report and review of the literature

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Necrotizing sialometaplasia is a self-limiting, variably ulcerated benign process affecting minor salivary glands. Accurate histological diagnosis is paramount, as it has been mistaken for malignancy, which has resulted in excessively aggressive and unnecessary radical surgery. A unique case of bilateral necrotizing sialometaplasia, presenting with anaesthesia of the greater palatine nerves, is described. An attempt at active therapy with intralesional steroids had no effect on the course of the condition.

Necrotizing sialometaplasia was first described in 1973 by Abrams and Melrose¹ with a number of similar cases reported the following year by Dunlap and Barker.² It is a self-limiting, variably ulcerated, benign process, affecting minor salivary glands. It occurs most commonly in the palate, although it has been reported in all sites where salivary gland tissue is located, including the nasal cavity sinuses, lower lip, tongue, cheek, retromolar pad, soft palate and larynx.²⁻⁶ The condition is significant because it may mimic squamous cell carcinoma or mucoepidermoid carcinoma, both clinically and histologically. Misdiagnosis has resulted in many cases of inappropriate radical surgery.⁶⁻⁸

The great variety of both clinical and

histological appearances of necrotizing sialometaplasia may be due to the different stages that this condition may undergo from initiation to healing of the lesion. Anneroth and Hansen⁹ stated that the following five stages occur in most cases: infarction, sequestration, ulceration, repair and healing. A sub-acute variant has been described.¹⁰ In the largest reported series, consisting of 69 cases, Brannon⁶ observed a mean age incidence of 46 years, and a predominance of 2:1 male to female and 5:1 white to African-Americans.

Necrotizing sialometaplasia presents in the majority of cases as a painful ulcerated lesion, however the symptoms and clinical appearance may vary.^{11,12} Antecedent swelling is not uncommon. The lesions are usually unilateral but bilateral lesions occur in as many as 20% of cases.⁶ Localised paraesthesia and pain referral to the ear and pharynx are variable features, which have been reported.^{6,13} Anaesthesia is a rare presentation.¹⁴ Complete healing occurs, without treatment, usually within 3 to 12 weeks.⁶

An unusual feature of this case report was bilateral anaesthesia in the distribution of the greater palatine nerves, which preceded and succeeded the appearance of the

typical swelling and ulceration of necrotizing sialometaplasia. This clinical picture may contribute to the misdiagnosis of necrotizing sialometaplasia as a malignant neoplasm.

CASE REPORT

A 30-year-old female was referred to the Department of Oral Surgery, Oral Medicine and Oral Pathology, at the Dublin Dental Hospital by her general dental practitioner. She presented with a 12-day history of bilateral palatal swellings and numbness of her palate, with associated difficulty in swallowing. Three weeks previously she had had an influenza-like illness and had been prescribed a course of phenoxymethylpenicillin. She had also experienced mild headaches, photophobia and tinnitus. Subsequently she developed bilateral paraesthesia and then complete anaesthesia in the distribution of both greater palatine nerves, 1 week before the appearance of bilateral palatal swellings. Pain was not a feature. Her medical history was non-contributory and she was a non-smoker.

Clinical examination revealed bilateral swellings at the junction of the hard and soft palate of similar size (Fig. 1), accompanied by objective, complete anaesthesia

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Fig. 1
Bilateral
palatal
swellings at
presentation

bilaterally in the distribution of the greater palatine nerves. An occipitomental radiograph showed that both maxillary antra were cloudy but an orthopantomogram showed no significant dental abnormality.

The palatal lesions were biopsied and doxycycline (200 mg stat, 100 mg od) was prescribed for five days whilst awaiting the biopsy report. On review five days later there was little change. Swabs taken from the lesion grew only commensal organisms. The biopsy was consistent with necrotizing sialometaplasia.

Microscopically, there was mild acanthosis and parakeratosis of the stratified squamous epithelium. The lobular architecture of the underlying minor salivary gland tissue was maintained. However partial infarction and extensive squamous metaplasia of the salivary ducts and acini, with prominent stromal mucin, was present. There was a diffuse mixed inflammatory infiltrate, consisting of neutrophils, plasma cells, occasional eosinophils, and foamy macrophages (Figs 2 and 3).

Clinical Course

Since necrotizing sialometaplasia is known to have an inflammatory component to the disorder and there is no reference to active therapy in the literature, this case presented a unique opportunity for an internally controlled evaluation of intra-lesional corticosteroid therapy on the rate of healing of the lesion and rate of recovery of the anaesthesia. With the patient's informed consent, 10mg of triamcinolone was injected into the swelling on the right side of the palate on three occasions at weekly intervals. The swelling on the left side was not treated and acted as the control.

After one month there was no difference in the rate of healing of the treated and untreated lesions or, unfortunately, the symptomatic anaesthesia. At this stage both swellings had completely resolved (Fig. 4). The anaesthesia persisted, gradually resolving over 18 months. There was no apparent effect of the corticosteroid therapy on the recovery from anaesthesia.

Aetiopathogenesis

Although the aetiopathogenesis of necrotizing sialometaplasia remains unknown, there is general consensus that an ischaemic event in the salivary gland precedes the development of the lesion. This is supported by the observation that ligation of the arterial supply to major salivary glands in rodents may result in a similar histopathological picture.^{15,16} The disease is also seen in sickle cell disease, which can cause infarction and the vasculopathies, Buerger's disease and Raynaud's disease or phenomenon; conditions which both predispose to ischaemia.^{17,18} Other ischaemic predisposing factors include: vascular damage due to trauma from intubation¹⁹ or fellatio,⁵ local anaesthetic needles,⁶ or chronic vomiting.²⁰ Addition of a

vasoconstrictor to local anaesthetic solutions may also play a part.²¹ Local radiotherapy,²¹ cocaine use², smoking (and alcohol)⁹ and pressure from local space-occupying lesions^{22,23} have also been implicated.

There is also an association with preceding upper respiratory tract infection within the previous few weeks.⁶ It is possible that the ischaemic event in these cases is due to immune complex disease, similar to the aetiology of erythema multiforme or benign trigeminal sensory neuropathy.²⁴

Histopathology

The entity known as necrotizing sialometaplasia comprises ischaemic lobular necrosis of sero-mucinous glands, but with maintenance of intact lobular architecture, despite coagulative necrosis of the mucinous acini.¹ Pale outlines of the acini often persist, but the nuclei are hypochromatic or absent. Extension of pools of mucin into the adjacent tissues elicits an inflammatory reaction dominated by histiocytes and granulation tissue. The inflammatory component within the necrotic lobules is often minimal, but is usually prominent in the surrounding tissues. Although squamous metaplasia of ducts and acini is a feature, the metaplastic cells have benign nuclear morphology, with minimal pleomorphism or hyperchromatism and few mitotic figures. The nests of squamous epithelium usually have a smooth periphery, however,

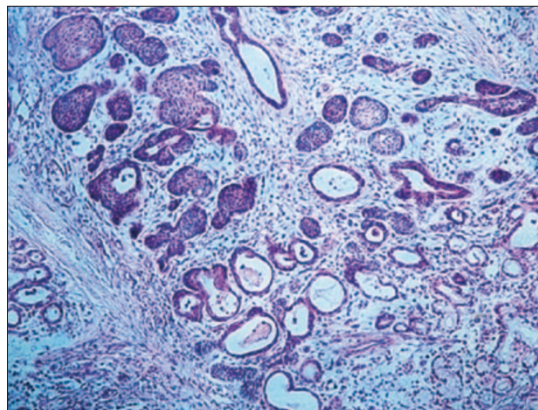


Fig. 2
Squamous
metaplasia of
ducts

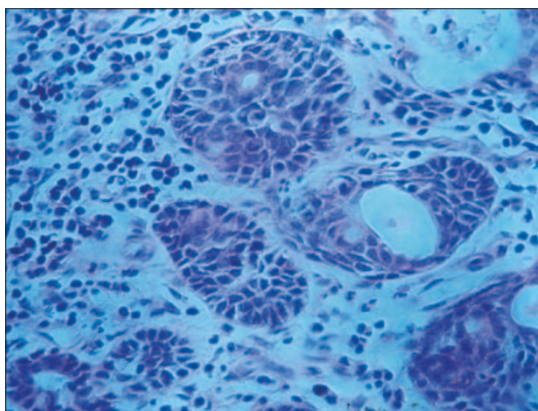


Fig. 3
Mixed
inflammatory
exudate

they occasionally have an irregular outline. The overlying or adjacent epithelium is often markedly hyperplastic with thick elongated and complex rete processes. This pseudo-epitheliomatous hyperplasia, along with extensive ductal metaplasia, may resemble squamous cell carcinoma.

It has been reported that histologic features may have some relation to the age of the lesion. Coagulative necrosis is a more dominant feature in the early lesions, whereas fibrosis and squamous metaplasia are features of an older lesion.⁶

DISCUSSION

This case represents, to our knowledge, the first documented case of bilateral greater palatine nerve anaesthesia, immediately preceding and succeeding the lesions of necrotizing sialometaplasia, and the first attempt at management by active therapy. Although the condition has a number of predisposing factors, the common outcome appears to be infarction of salivary glands or the sero-mucinous glands of the upper respiratory tract caused by a compromised blood supply due to vascular injury of different kinds. These lesions had an aggressive appearance suggestive of neoplasia including features such as a rapidly growing mass with associated anaesthesia. The bilaterality and histological features, and ultimately the clinical course, however, suggested otherwise. The presence histologically of lobules of infarcted minor salivary glands, along with marked squamous metaplasia that remains cytologically bland, suggested a diagnosis of necrotizing sialometaplasia.

Necrotizing sialometaplasia may occur *de novo*, but may also be seen in association with other space-occupying lesions, both benign or malignant.²⁵ Because of the latter, whenever the diagnosis of necrotizing sialometaplasia is made, close follow up is indicated until healing is complete. Recognition of the histological profile, and the varied clinical settings in which necrotizing sialometaplasia can be found, is essential, to avoid histopathological misinterpretation and inappropriate treatment for this benign reactive condition.

Since, in this case, there was a preceding upper respiratory tract infection with clinical and radiographic evidence of persistent sinusitis, we postulated that, following treatment of any sinus infection, the lesion might respond to intra-lesional anti-inflammatory therapy. The vasculopathy might be a late manifestation of immune complex disease (in this case following the

upper respiratory tract infection), analogous to the suggested pathogenesis of erythema multiforme and benign trigeminal sensory neuropathy.²⁴ The neuropathy is presumably due to the ischaemic process affecting the vasa nervorum of the greater palatine nerves.

The fact that the lesions were bilateral, of similar size, and there was also bilateral anaesthesia allowed the opportunity to evaluate the effect of intralesional steroid treatment on the condition, using the contra-lateral lesion as a control. The usual management is simple observation until the healing phase is complete. We are unaware of any previous reports of active therapy. The lack of response to this treatment is disappointing and probably reflects the fact that the necrotizing process was already complete before the treatment was started. Instituting the treatment earlier would be difficult clinically, since there is always a delay because the diagnosis is histopathological.



Fig. 4 Complete resolution of both palatal swellings after 1 month.

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