

## View from Beneath: Pathology in Focus

### Necrotizing sialometaplasia

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#### Abstract

Necrotizing sialometaplasia is a benign self-limiting disorder which can simulate malignancy. We report the third such case of nasal fossa involvement in the literature. The pathology of this condition is discussed.

#### Introduction

Necrotizing sialometaplasia is an uncommon but important condition. It was first described by Abrams *et al.* (1973). Initially it was thought to involve only the hard palate, but was subsequently described in the parotid gland, (Donath, 1979; Batsakis and Manning, 1987), retromolar pad (Forney *et al.*, 1977), larynx (Walker *et al.*, 1982), and nasal cavity (Chen, 1982; Close and Cowan, 1985).

Since 1973, about 80 cases have been reported, of which only two involved the nasal fossa.

Necrotizing sialometaplasia is an inflammatory disorder which principally affects minor salivary glands (Maisel *et al.*, 1977), but can also affect major glands (Batsakis and Manning, 1987; Seifert, 1991). Clinically and microscopically it may simulate squamous or mucoepidermoid carcinoma, but is pathologically benign and self-limiting (Batsakis and Manning, 1987).

Abrams (1986) has listed five histopathological features which are most helpful in distinguishing it from malignancy. He also suggests that this entity be reserved for those cases where spontaneous infarction of the gland has occurred.

The purpose of this report is to present the third case of necrotizing sialometaplasia of the nasal fossa. It is intended to remind and alert pathologists of this benign and self-limiting disorder which can simulate malignancy.

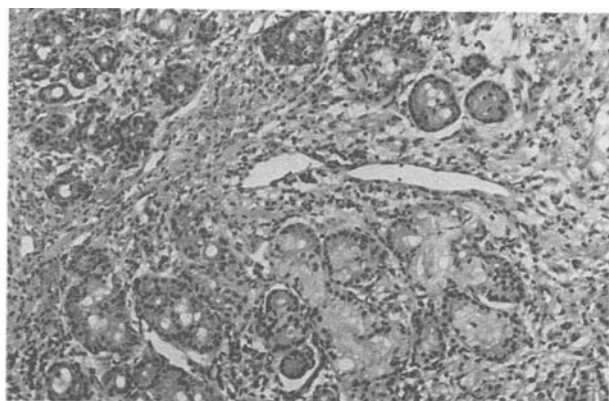


Fig. 1

Showing the characteristic ulcerated surface of the lesion. Note the atrophic lobulated glandular tissue separated by fibrous septa. There is no sign of metaplasia in these glands.

#### Case report

A 50-year-old man was admitted with a two-day history of recurrent right-sided epistaxis. Examination revealed a bleeding area on the right anterior septum which was cauterized with silver nitrate. Epistaxis recurred and a BIPP pack was inserted for 24 h. The bleeding recurred after the pack was removed and it was decided to examine the nose under general anaesthesia.

An irregular polypoidal mass on the right inferior turbinate was noted. This was biopsied, the specimen sent for histology and a further anterior pack inserted. Initial histopathological examination suggested a muco-epidermoid carcinoma but this diagnosis was revised to necrotizing sialometaplasia on review. At two years follow-up, there have been no further episodes of bleeding and the lesion has resolved completely.

#### Pathological findings

Microscopy showed the characteristic ulcerated nasal mucosa, below which there was some atrophic glandular tissue divided by fibrous tissue and displaying the characteristic lobular pattern (Fig. 1). Squamous metaplasia was variable but the glands deeper in the fibrotic mucosa showed considerable squamous metaplasia of larger and smaller groups of acini (Figs. 2–4). The small ducts were dilated and contained a good deal of PAS-positive mucus secretion.

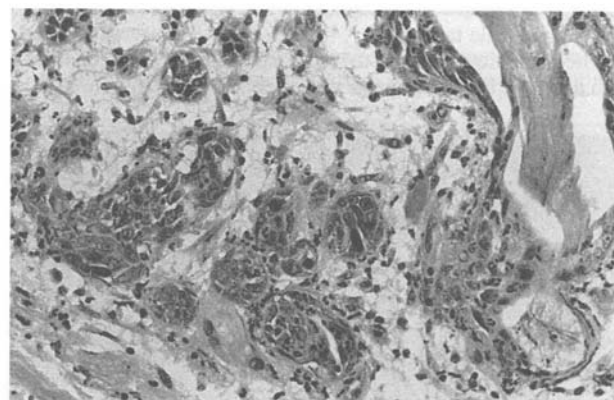


Fig. 2

A deeper lobule composed of acini shows extensive squamous metaplasia confined to the acini. There is a dilated duct present containing much PAS-positive secretion. There is no evidence of invasion.

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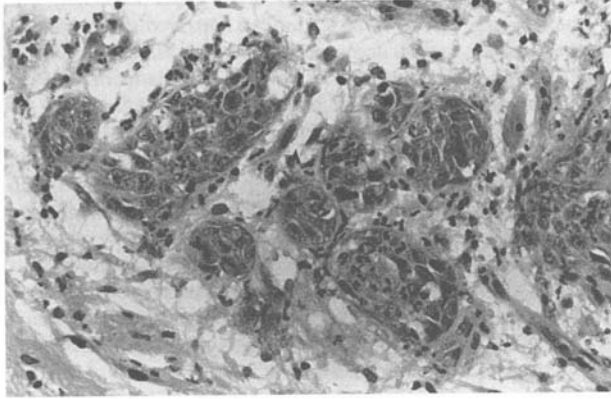


Fig. 3

Showing the cytological detail of the squamous epithelium. Note marked irregularity of the nuclear size and shape and hyperchromasia. No mitoses were seen.

At higher magnification, some irregularity of nuclear size and shape was noted and there was considerable hyperchromasia of the nuclei. These were the microscopical features regarded with some suspicion at first inspection and appeared to warrant the tentative diagnosis of a mucoepidermoid carcinoma (Figs. 2 & 3).

There was, however, no evidence of invasion and the squamous metaplasia was contained with occasional exception within the acini (Fig. 4). There was no disruption of the lobular pattern of the small salivary glands. An important finding was the presence of small occluded vessels within the fibrous tissue surrounding the glands. (Figs. 5 & 6). The term 'salivary gland infarction' has been suggested in the new classification (WHO) of salivary gland tumours because the lesion resembled infarcts in some other organs (Seifert, 1991).

The tentative diagnosis was subsequently revised to that of necrotizing sialometaplasia and the favourable clinical progress has confirmed the histopathological diagnosis.

**Discussion**

Necrotizing sialometaplasia was first described by Abrams *et al.* in 1973, although it was first recognized by Cornyn.

It is a benign, inflammatory, self-limiting disorder of unknown aetiology. The disease affects salivary gland tissue, particularly the minor salivary glands of the hard palate (Walker *et al.*, 1981). About 80 cases have been reported so far (Chen, 1982), and it is now realized that any site with salivary gland tissue is a possible target.

The condition appears to be more common in males than females, and is not confined to Caucasians. The age incidence

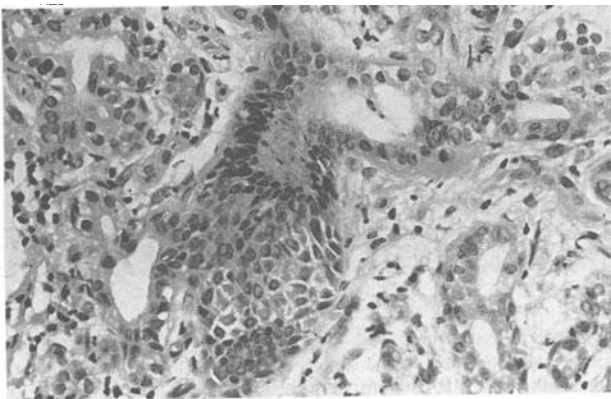


Fig. 4

Detail of acinus showing squamous metaplasia apparently beyond the confines of the acinus.

ranges between 18 and 83 years with a mean of 47 years; 73 per cent of the cases were over 40 years, (Gad *et al.*, 1980). Patients usually present with a painless or minimally painful ulcer. Only six cases have been described presenting as a mass or swelling (Close and Cowan, 1983).

Abrams *et al.* in 1973 described five pathological features which are helpful in distinguishing necrotizing sialometaplasia from other more serious lesions:

- 1) Lobular infarction or necrosis.
- 2) Bland appearing nuclear morphology of the squamous cells.
- 3) Simultaneous metaplasia of ducts and mucous acini.
- 4) Prominent granulation tissue and inflammatory components.
- 5) Maintenance of the general lobular morphology in spite of the extensive inflammatory and metaplastic changes involving more than one lobule.

It may be of interest that electron microscopic studies of tissues removed from a case of necrotizing sialometaplasia of the palate (kindly provided by Professor W. Arnold, Luzern) showed only goblet cells filled with secretory globules and loosely arranged squamous epithelial cells probably of metaplastic origin. There were no unusual features of assistance to the diagnosis.

There is a general agreement with the view that the essential histological basis is an infarct of the salivary gland lobules with subsequent repair and metaplasia. However, the pathogenesis of the condition has remained unclear. Several mechanisms have been proposed. Among these are arteriosclerosis, chronic irritation secondary to consumption of tobacco and alcohol, previous surgery and acute trauma. The most widely believed, however, is ischaemia of the salivary gland tissue as indicated also by the finding of occluded vessels in the nasal biopsy from our patient.

Englander and Cataldo (1976) and Standish and Shafer

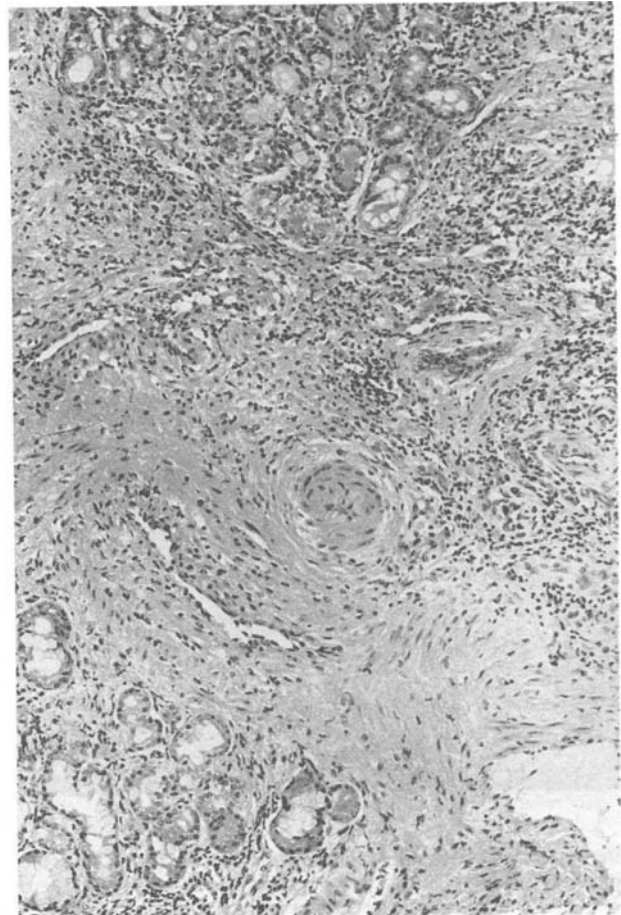


Fig. 5

Note small occluded artery in the surrounding dense fibrous tissue. (consistent with the description as 'salivary gland infarction').



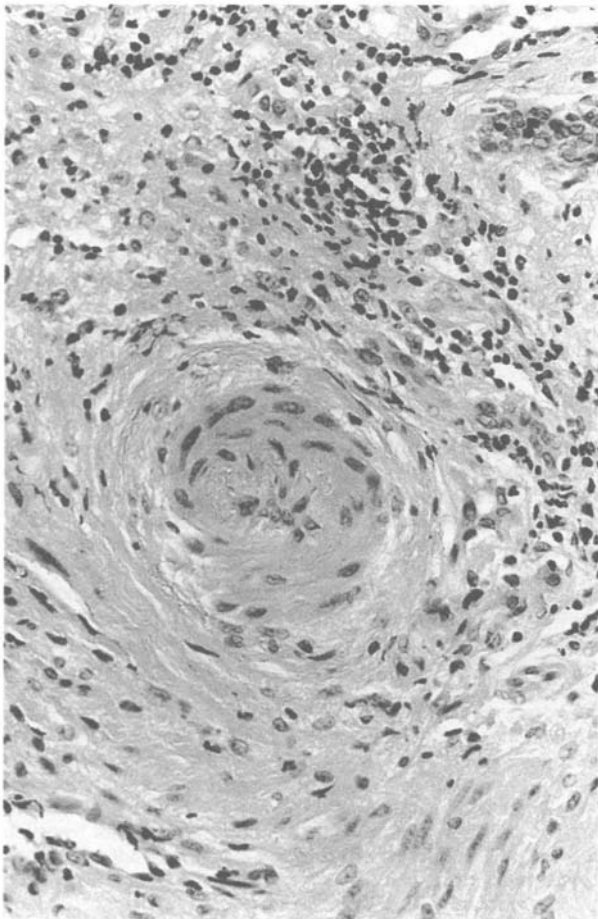


Fig. 6

Detail of occluded vessel probably causing the surrounding atrophy of the surrounding minor salivary glandular tissue partly replaced by dense fibrotic tissue.

(1957), produced similar lesions in rats after ligation of the ducts and arteries of major salivary glands. Walker *et al.* (1981) described a case of necrotizing sialometaplasia of the larynx secondary to atheromatous embolization. Johnston (1977) described a case of sialometaplasia occurring in a patient 10 days after radical maxillectomy in the residual mucosa. These changes, he believes, were the direct result of impairment of the vascularity of the affected mucous glands at operation.

Batsakis and Manning (1987) have described eight cases occurring in major salivary glands (parotid). It is interesting to note that 'seven of the eight lesions followed an operative procedure on the parotid and presented within a period of three and half weeks after the initial surgery'. Also five of Donath's six cases followed a surgical procedure on the parotid gland (Donath, 1979).

Necrotizing sialometaplasia is an important disease because of its ability to mimic malignancy. The importance of reaching a correct diagnosis cannot be overstated. Our patient could have been subjected to further surgery on the basis of the first histopathological report. There have been several cases in the literature of unnecessary radical surgery. Maisel *et al.* (1977) described a case of a 68-year-old woman who had a partial maxillectomy for presumed squamous carcinoma on nasal biopsy. Histology of the specimen failed to reveal carcinoma, but instead features suggestive of necrotizing sialometaplasia. The unwary pathologist unfamiliar with this entity could be misled by the combination of mucous cells and proliferating, keratinizing

squamous epithelium, to suggest a diagnosis of mucoepidermoid or squamous carcinoma (Gad *et al.*, 1979). The primary histological diagnosis in the eight cases reported by Batsakis and Manning (1987) was with one exception of a familial neoplasm encountered in the parotid gland.

The treatment of this condition has varied in the past from wide resection to local excision biopsy. A simple biopsy is all that is required as all cases resolve spontaneously within three months (Close and Cowan, 1983). There have been no cases in the literature of recurrence at follow up periods of up to eight years.

It is interesting to speculate that the long survivals reported for low grade mucoepidermoid carcinomas may in fact reflect incorrect diagnoses in cases of necrotizing sialometaplasia (Maisel *et al.*, 1977).

Our patient presented without any previous surgery to his nose and clinical progress following the removal of the lesion was entirely uneventful confirming the diagnosis of necrotizing sialometaplasia.

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