CASE REPORT

CT findings of necrotizing sialometaplasia

A Suomalainen^{*1}, J Törnwall² and J Hagström³

¹Department of Radiology, Helsinki University Central Hospital, Helsinki, Finland; ²Department of Oral and Maxillofacial Surgery, Helsinki University Central Hospital, Helsinki, Finland; ³Oral Pathology Unit/HUSLAB, Helsinki University Central Hospital, Helsinki, Finland

An otherwise healthy 56-year-old Caucasian female was referred to the radiology department because of an ulceration of her palatal mucosa. Clinically the lesion was suspected to be malignant and a multislice CT examination of the head and neck region was performed. This revealed an ulcer-like cavity with no tumour-like contrast enhancement. No relevant bone changes or suspicious lymph nodes of the neck were detected. Based on the multislice CT findings, necrotizing sialometaplasia was suspected and the diagnosis was verified histopathologically. This report describes the CT findings of necrotizing sialometaplasia at the ulceration stage.

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Introduction

Necrotizing sialometaplasia (NS) is a benign self-limiting inflammatory condition which can be found at any site in the body that contains elements of the salivary glands,^{1,2} the most common site being the palatal mucosa.³ NS can be confused clinically and histologically with carcinoma, especially with mucoepidermoid carcinoma and squamous cell carcinoma.^{3–6} Granulomatous diseases such as syphilitic gumma and deep fungal infections can also mimic NS.⁷ The cause of NS is uncertain. Most authors believe it to be the result of ischaemia of the salivary tissue which leads to a local infarction. Because NS mimics a malignant process both clinically and microscopically, and can be mistaken for a malignancy, its recognition is important.

Histologically, necrotizing sialometaplasia is characterized by acinar necrosis in early lesions with concomitant squamous metaplasia of the salivary ducts as the condition progresses. Despite the necrosis, the lobular architecture of the glands involved is preserved.⁸ The extent of squamous metaplasia may be considerable and may even mimic a malignant lesion. Imaging findings in cases of NS have been reported only very rarely. MRI findings of NS have been reported recently by Farina et al.⁹ Lee et al.¹⁰ reported

MRI findings of NS accompanied by adenoid cystic carcinoma on the soft palate.

Case report

A 56-year-old Caucasian female in good general health was admitted to the Department of Oral and Maxillofacial Surgery of Helsinki University Central Hospital, Finland. The patient, who had a partial denture in her maxilla, had noticed ulceration of her palatal mucosa 4 weeks earlier (Figure 1). The ulceration was asymptomatic and located at the junction of the hard and soft palates, left of the midline. Clinically the lesion was suspected to be malignant and a multislice CT (MSCT) (LightSpeedTM; General Electric Medical Systems, Milwaukee, WI) examination of the head and neck region was performed. The MSCT examination was performed first without contrast medium in the palatine region followed by contrast enhancement from the skull base to the clavicle [Ultravist[®], 370 mg I/ml; Schering, Berlin, Germany (100 ml)]. The MSCT examination revealed an ulcer-like cavity, but with no tumour-like contrast enhancement of the lesion (Figure 2). No relevant bone changes (Figure 3) or suspicious lymph nodes of the neck region were detected. Based on the MSCT findings, NS was suspected. An incisional biopsy sample of the lesion was obtained after the MSCT

*Correspondence to: Dr Anni Suomalainen, Department of Radiology, Helsinki University Central Hospital, PO Box 263 (Kasarmikatu 11–13), FI-00029 HUS, Finland; E-mail: anni.suomalainen@hus.fi
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Figure 1 Clinical photograph showing ulceration at the junction of the hard and soft palates, left of the midline

examination and the NS diagnosis was verified histopathologically.

Histologically the mucosal epithelium displayed pseudoepitheliomatous hyperplasia. Acinar degeneration in the salivary glands was detected together with ductal squamous cell metaplasia of a different volume (Figure 4).

1 week after the biopsy, the lesion of the palatine ulcer was unchanged, but after 1 month the lesion was completely healed and follow-up was discontinued.

Discussion

NS was first described by Abrams *et al*⁴ in 1973. Most patients are older than 40 years, although there is a wide age range at presentation.^{3,11} The incidence appears to be twice as high in males as in females.³ NS appears initially as a non-ulcerated swelling,

Figure 3 Multislice CT examination revealed no bone changes (sagittal view, bone window)

possibly associated with pain. Within 2–3 weeks, necrotic tissue typically sloughs out, leaving a crater-like ulcer.⁸ Complete remission occurs without treatment in 4–90 days.⁶ The pathogenesis of NS has been defined by five histological stages: infarction, sequestration, ulceration, reparative stage and healed stage.¹² However, these changes can overlap and depend on the extent and severity of damage.¹² Our patient had noticed the palatal lesion 4 weeks before the CT examination was performed. The CT examination showed an ulcer-like defect reflecting the common healing process and the ulcer had spontaneously healed completely 1 month after the CT examination.

Farina *et al*⁹ reported a case of MRI findings of NS. They detected a lobulated submucosal lesion in the palate that was hyperintense on T_2 weighted images and hypointense on T_1 weighted images. Only a thin peripheral rim enhancement was observed after contrast administration. They concluded that a fluid-like appearance of a lesion and the absence of contrast enhancement could help to exclude salivary gland neoplasm and that these might be the MRI findings suggestive of NS. In their case, the MRI scan was performed 2 weeks after the patient had noticed the palatal swelling. The differences between our CT findings and their MR findings can be explained by the natural self-healing process of NS; first the

Figure 2 Contrast-enhanced multislice CT examination revealed an ulcer-like cavity with no tumour-like contrast enhancement of the lesion in the (a) axial and (b) coronal views

Figure 4 Necrotizing sialometaplasia with (a) little squamous metaplasia (arrows) and with (b) intense metaplasia (arrows). Arrowheads indicates acinar structures

submucosal lesion with peripheral inflammatory reaction followed by the ulcer-like defect. Farina *et al*⁹ speculated that the peripheral rim enhancement might represent the inflammatory reaction. The degree of the inflammatory reaction can be expected to vary depending on the stage of the NS and from one case to another, as stated by Anneroth and Hansen.¹² Also, MR images can show minor rim enhancement more clearly than CT images. In addition, biopsy of the lesion before imaging may affect the imaging findings. In the case reported by Farina *et al*⁹, a biopsy of the lesion had already been performed before the MR examination. In our case, the incisional biopsy specimen was obtained after the MSCT examination. Bone involvement of the hard palate was not noted in either of these cases. This is in accordance with the literature. Although bone involvement is an infrequent finding, it does not preclude the diagnosis of NS.³

However, the natural cause of NS can vary. It has been reported that necrosis may not feature in the presentation of sialometaplasia in young children.¹ Ylikontiola *et al*¹ reported a case of sialometaplasia of the soft palate in a 2-year-old girl after adenoidectomy. Frank necrosis was not evident from a clinical and histological examination in her case. Imbery and Edwards⁶ stated that if the infarct

is limited in extent, sequestration and ulceration do not occur. Ylikontiola *et al*¹ speculated that the adenoidectomy could have caused the infarction of the salivary gland tissue in their patient. Our patient had a maxillary partial denture, which has been reported to be one predisposing factor for NS.^{3,4} In addition to prior surgery and local trauma or injury, other predisposing factors include dental injections, alcohol abuse, smoking, cocaine use, upper respiratory infections, radiation and adjacent tumour growths.^{2,3,13} Bulimia may also be an underlying cause of NS.^{2,11}

Even though adjacent tumour growths have been reported as predisposing factors for NS, the association of NS with neoplastic conditions is a relatively rare finding.³ Brannon *et al*³ reported 8 cases of NS associated with adjacent neoplasm or a cystic lesion out of 184 cases, with 4 of them occurring on the palate. A case of NS accompanied by adenoid cystic carcinoma on the soft palate together with MRI findings has been reported quite recently.¹⁰ These authors concluded that the MRI findings might be different for each stage of NS. In their case, neither a fluid-like appearance nor the absence of contrast enhancement was found in MRI. On the contrary, the gadolinium-enhanced T_1 weighted images showed diffuse enhancement. For this

reason a wide mass was surgically removed even though the final histopathological report showed that adenoid cystic carcinoma was present in only a small portion of the mass. It seems to be difficult to differentiate NS from a tumour when it is accompanied by a malignancy.

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To conclude, we have described the CT findings of NS at the ulceration stage. Imaging findings of NS seem to vary from stage to stage, as would be expected. Further studies are needed to evaluate the common CT and MRI findings of NS at different stages.