CASE REPORT

Imaging findings of necrotizing sialometaplasia of the parotid gland: case report and literature review

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Although necrotizing sialometaplasia (NS) of the parotid gland is rare and occasionally presents as a lesion that mimics a malignant tumour, imaging findings in cases of NS have been rarely reported. We describe here a case of NS in which there was an increasing lesion manifesting overnight on the parotid gland in an 83-year-old male. We also investigated the use of pre-operative imaging based on previous reports and discuss the importance of these images in helping to guard against overzealous treatment. It is critically important to closely examine whether there are aspects of NS, such as the present case, in pre-operative MRI findings that can be useful in proper diagnosis and treatment.

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Introduction

Necrotizing sialometaplasia (NS) is a benign inflammatory disease often found in the minor salivary gland and seldom observed in major salivary glands.¹ To the best of our knowledge, the number of imaging reports associated with NS arising from the parotid gland is very small. In some of the cases, unnecessary surgical excision and inappropriate therapy were performed because of misdiagnosis, using an ostensible definitive diagnosis based on pathological findings.^{2,3} Although the pathogenesis of lesions is thought to be owing to ischaemic changes in the salivary glands arising from causes such as traumatic injury and medical procedures, the precise aetiology has not been fully elucidated.^{2,4} We report here a case of NS in the parotid gland, with the definitive diagnosis confirmed after superficial parotidectomy. Furthermore, we discuss the importance of pre-operative imaging to guard against unnecessary treatment.

Case report

An 83-year-old male was referred, complaining of an acute enlarging mass in the right parotid gland that had developed overnight. The patient had been taking oral medications for hypertension and hyperlipaemia for several years. There was no history of trauma or surgical procedure of the parotid gland. On physical examination, a palpably hard and firm mass, measuring 40 × 50 mm, was found on the right parotid gland tail (**Figure 1**). Additionally, the patient had a haemorrhagic macule on the forearm. There was no discharge of pus from the parotid duct.

Ultrasonographic image (linear probe PLT-1204BT, 13–18 MHz, Xario XG; Toshiba, Odawara, Japan) of the mass had a inhomogeneous hypoechoic appearance in comparison with a normal parenchyma of the parotid gland. The size of the mass was 26.5 × 21.8 × 19.9 mm, and the border was unclear (**Figure 2**). CT images (Acquilion ONE™; Toshiba) revealed a slightly high density mass in the parotid

Figure 1 A palpable hard and firm mass was found on the right parotid gland tail at the first visit.

gland. The border between the mass and the normal parotid gland tissue was unclear, suggesting the inflammatory mass in the parotid gland tail (Figure 3). On MRI (Signa HDxt 1.5T; GE Healthcare, Tokyo, Japan), T_2 weighted image (WI) revealed an apparently well-circumscribed mass of 26 mm that was surrounded with high signal intensity. The outline of the wall of the mass was observed as low signal on both T_1 -WI and T_2 -WI, suggesting that the lesion was covered with a capsule. The content of the mass exhibited a low intensity

area as well as an asteroid high intensity area on T_2 -WI (Figure 4a,b). The slightly high intensity on the inner wall of the mass on T_1 -WI was observed (Figure 4c). This area was also observed as a high intensity area on T_2 -WI. These findings suggested that degeneration and necrosis had occurred within the mass.

To rule out a malignant tumour, we performed gallium scintigraphy (^{67}Ga ; Symbia™ E; Toshiba) as an auxiliary diagnosis. A faint accumulation of gallium was observed over the whole area of the right parotid gland in comparison with the left parotid gland, indicating that the lesion might not be owing to a malignant tumour but to inflammation (Figure 5).

We suspected a tumour-like lesion displaying NS with surrounding inflammation. To reach definitive diagnosis, superficial parotidectomy with certain peripheral tissues around the mass was performed after extinction. Microscopic findings after haematoxylin and eosin staining of the section exhibited a mixture of normal salivary gland tissue (Figure 6a), squamous metaplastic tissue conforming to the outline of salivary ducts without any atypia (Figure 6b) and necrotic tissue (Figure 6c), consistent with NS. After 6 months of follow-up, no recurrence has developed.

Discussion

NS, an inflammatory reaction of salivary tissues with a self-limiting disease course, was described in 1973 by Abrams et al.² It mainly occurs in the minor salivary glands of the hard palate. The incidence of this lesion in the parotid gland is rare and has been estimated to be at most approximately 10%.³ To our knowledge, Donath⁵ reported six cases of NS occurring in the parotid gland

Figure 2 Ultrasonography showing the inhomogeneous hypoechoic appearance on the right parotid gland. The size of the mass was (a) 26.5 mm × (b) 21.8 mm × (c) 19.9 mm.

Figure 3 CT images showing the poorly margined mass with normal parotid gland tissue.

in 1979, following which Batsakis and Manning¹ documented an additional seven cases with clinical and pathological findings in 1987. Furthermore, Brannon *et al*³ reviewed 69 cases of NS, including 6 cases in the parotid gland in 1991. As far as we have been able to determine from the English literature, excluding NS accompanied by salivary tumour, such as the metaplastic Warthin's tumour,⁶ five reports^{4,7-9} of NS of the parotid gland have appeared, including the current case.

An examination of the five reports on NS in the parotid gland showed that the age ranged from 17 to 83 years, with an average age of 53.4 years. Clinical symptoms most commonly comprised swelling of the neck, followed by pain (Case 20), discharge of pus from the salivary duct (Case 21) and swelling of the pharynx and neck, vocal cord paralysis and neck lymphadenopathy (Case 22) (Table 1). Most of the masses was <20 mm, consistent with previous investigation.¹ In the present case, the size of the lesions was larger than the average size in previous reports.¹

The aetiology of NS remains unknown, but it may be associated with salivary gland tissue ischaemia,³ leading to infarction and subsequent necrosis of the tissue, followed by repair and metaplasia. In 1987, Batsakis and

Figure 5 A faint accumulation of gallium-67 was observed over the whole area of the right parotid gland in comparison with the left parotid gland.

Manning¹ reported that seven of eight parotid gland cases had followed an operative procedure for another primary tumour of the parotid gland. It is noteworthy that the clinical manifestations presented within a period of 3.5 weeks after the initial surgery. Since 2002, the number of reasons for operation of the parotid glands has decreased owing to the following histological criteria. The other causes were thought to be pressure-induced ischaemia,⁷ vascular injury^{4,9} and tumour.⁸ In the present case, it was speculated that the cause was owing to vascular fragility as indicated by the haemorrhagic macule on the forearm, similar to the mechanical pressure as the aetiological factor leading to ischaemia reported by Prabhakaran *et al*.⁷

As shown in Table 1, our investigation demonstrated that ultrasonography, CT and fine needle aspiration biopsy were performed in most cases for pre-operative diagnosis. Although MRI findings of NS of the parotid gland have never been reported, Farina *et al*¹⁰ reported a case of MRI findings of NS manifesting ulceration of the palatal mucosa. They speculated that the high intensity on T_2 -WI might be the feature of salivary gland necrosis, and the peripheral rim enhancement might reflect the inflammatory reaction, leading the MRI findings to be suggestive of NS. In the present case, the high intensity on the inner wall of the mass on T_1 -WI

Figure 4 (a) An axial section and (b) a coronal section with T_2 weighted MRI showing an apparently well-circumscribed mass with surrounding high signal intensity in the right parotid gland tail. (c) A coronal section with T_1 weighted MRI showing high intensity on the inner wall of the mass.

Figure 6 (a) Normal salivary gland tissues can be seen in the right portion, and the inflammatory tissues are present in the left portion. (b) The fibrous connective tissue with inflammatory cell infiltrate and squamous metaplastic tissues conforming to the outline of the salivary ducts can be observed. (c) On a high-power view, squamous metaplasia without cytological atypia around the salivary gland duct can be observed. (d) Necrotic tissues and inflammatory infiltration with histiocytes can be observed. Original magnifications: (a, b) $\times 40$; (c) $\times 200$; (d) $\times 100$, haematoxylin and eosin stained.

and an asteroid high intensity area within the mass on T_2 -WI were observed, suggesting that tissue degeneration and necrosis had occurred within the mass. The well-circumscribed mass was surrounded with high intensity signal on T_2 -WI, indicating that the inflammation may have spread peripherally. Furthermore, gallium scintigraphy supported the determination

that this was not a malignant tumour. These pre-operative findings indicated that the lesion in the parotid gland tail was most likely the result of a tumour-like lesion displaying NS accompanied by inflammation. Di Palma *et al*⁶ reported that trauma such as a fine needle aspiration biopsy caused infarction and squamous metaplasia in a case of Warthin's tumour,

Table 1 Characteristics of the reported cases of necrotizing sialometaplasia arising in the parotid gland

Case	Year	Author	Age (years)/sex	Cause	Size (cm)	Clinical presentation	Pre-operative examination
1–6, 7–13	1979, 1987	Donath ⁵ Batsakis and Manning ¹	Mean age of M: 54 Mean age of F: 49 Sex ratio: M:F = 1:2	Post-operative vascular injuries (11/13 cases)	0.6–1.0 (mean of six cases)	Post-operative salivary mass: 86% Sialadenitis: 14%	NM
14–19	1991	Brannon <i>et al</i> ³	NM	Post-operative vascular injuries (5/6 cases)	NM	NM	NM
20	2002	Aydin <i>et al</i> ⁴	17/F	Vascular injury	2	Pain Swelling of neck	Ultrasonography/CT/ FNAB
21	2006	Prabhakaran <i>et al</i> ⁷	32/M	Pressure-induced ischaemia	NM	Swelling of neck Pus discharge from the parotid duct	CT
22	2010	Yoshioka <i>et al</i> ⁸	66/M	Malignant lymphoma	NM	Swelling of neck Swelling of pharynx Vocal cord paralysis Neck lymphadenopathy	FNAB
23	2013	Kim <i>et al</i> ⁹	69/F	Vascular injury owing to heavy smoking	3 $\times$ 2, 1.5 $\times$ 1.5	Swelling of neck	CT/FNAB
24	2014	Current report	83/M	Pressure-induced ischaemia	4 $\times$ 5	Swelling of neck	Ultrasonography/CT/ MRI/Ga scintigraphy

F, female; FNAB, fine needle aspiration biopsy; Ga, gallium; M, male; NM, not mentioned.

following that a lesion analogous to NS was observed in non-neoplastic salivary glands. Scrupulous attention is required to perform fine needle aspiration biopsy in the course of an investigation of patients with a mass in the neck. Thus, we have stressed that MRI findings might be necessary and helpful for the proper diagnosis and treatment of NS.

However, this can present a diagnostic dilemma owing to histological findings as well as the clinical examination and imaging, all having the potential to mimic a malignant neoplasm, such as squamous cell carcinoma or mucoepidermoid carcinoma.^{2,3} To avoid needless surgery, certain reports have suggested the following specific criteria for pathological diagnosis: necrosis of salivary tissue; a time variable prominence of granulation and acute and chronic inflammation; squamous metaplasia conforming to duct and/or acinar outlines; and maintenance of the salivary morphology.¹⁻³

Anneroth and Hansen¹¹ classified the pathological condition into five stages, following infarction (necrotic), sequestration, ulceration, reparative and healed. Suomalainen *et al*¹² suggested that the degree of the inflammatory reaction can be reflected in the above stages of NS. In the present case, the specimen histologically

displayed variable features consistent with different stages, including the necrotic, reparative and healed, apparently because the period from the onset to operation was a relatively long 1 month.

In conclusion, NS is a spontaneously resolving inflammatory process of the salivary gland that can mimic a malignant tumour. It is critically important to closely examine whether there are aspects of NS in the pre-operative MR findings or not to avoid overzealous treatment. If the lesion in the parotid gland is not suspected of being a malignant tumour but rather an NS, it might be preferable to adopt a “wait and see” approach owing to the possibility of spontaneous healing within 3–12 weeks³ that may occur in cases of NS. When surgical excision of the lesion is performed, we should carefully examine whether there are aspects of NS in the histopathological findings, by following the pathological guidelines described above.

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