

## CASE REPORT

# Sialometaplasia arising in the ectopic salivary gland ductal inclusions of multiple intraparotid lymph nodes

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Sialometaplasia, squamous metaplasia of salivary gland lobules, is a benign condition occasionally presenting with lesions clinically simulating malignancy. "Necrotising sialometaplasia", recognised by lobular infarction, necrosis, and simultaneous squamous metaplasia of ducts and acini is a well known condition. There are only a few reports of the "proliferative type of sialometaplasia", which is recognised by a more mature morphology of larger and more irregular metaplastic nests, lacking necrosis. This report describes a unique case of "proliferative sialometaplasia of multiple intraparotid lymph nodes" occurring in a 55 year old woman, presenting with multiple parotid lumps. This interesting case points to the importance of intraparotid lymph nodes as sites for multiple lesions of the parotid region.

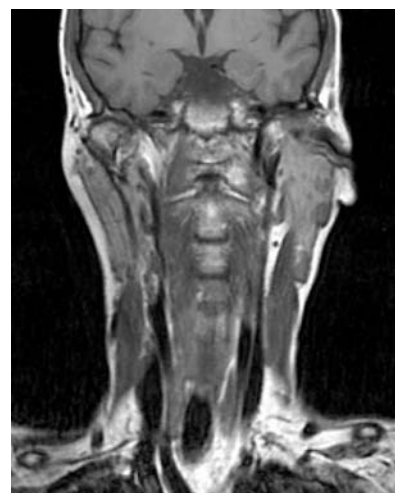
Sialometaplasia, squamous metaplasia (SM) of salivary gland lobules, is a benign condition occasionally presenting with lesions mimicking malignancy. Since the description of the necrotising type,<sup>1</sup> many cases have been published. The proliferative form has only been reported in a few cases in intraparotid lymph nodes (LNs).<sup>2,3</sup> This type of sialometaplasia is recognised by solid squamous metaplastic nests involving ductal structures and lacking atypia and necrosis.<sup>2</sup>

### CASE REPORT

A 55 year old woman presented with left infra-auricular multiple lumps, of two years duration. She had undergone stapedectomy of the right ear five years previously.

The patient had a 1.5 × 1.5 cm, non-tender, semimobile mass in the left preauricular region, and a similar mass in the tail of the left parotid gland. The functions of the facial nerve were bilaterally normal and the examination of the other head and neck sites including the other salivary glands was unremarkable. She had no complaints of xerostomia. Sonography revealed hypochoic parotid nodules. The lesions were hyperdense with contrast enhancement on computed tomography scans. Magnetic resonance imaging demonstrated two spherical discrete masses. Lesions were hypointense on the T1 images and isointense on the T2 images with contrast enhancement (fig 1). The radiological conclusion was consistent with a benign tumour, in particularly pleomorphic adenoma, and the clinical diagnosis was that of pleomorphic adenoma and associated lymphadenomegaly of intraparotid LNs. The routine laboratory examinations were not remarkable: white blood cell count,  $8.1 \times 10^9$ /litre; haemoglobin, 135 g/litre; haematocrit, 39.3%; erythrocyte sedimentation rate, 21 mm/hour; antinuclear antibodies, antineutrophil cytoplasmic antibodies, human immunodeficiency virus were negative.

Left superficial parotidectomy with preservation of the facial nerve was performed.



**Figure 1** Coronal T1 weighted image; hypointense circumscribed mass lesions in the left parotid gland.

### Pathological examination

Superficial parotidectomy revealed two encapsulated, 1.5 cm, grey/white nodules located at the poles. Histology showed that they were both LNs and six additional LNs were also identified. Five LNs revealed ductal salivary gland inclusions, almost totally replaced by SM, but retaining the lobular architecture of ductal inclusions (figs 2, 3). Ductal inclusions were also identified in another lymph node. The parotid gland showed atrophic changes.

The metaplastic squamous epithelium was keratin positive and mucicarmen negative. Smooth muscle actin and S-100 was positive at the periphery of the metaplastic lobules (fig 4). The diagnosis was that of "proliferative sialometaplasia (PS)" involving salivary gland inclusions of multiple intraparotid LNs.

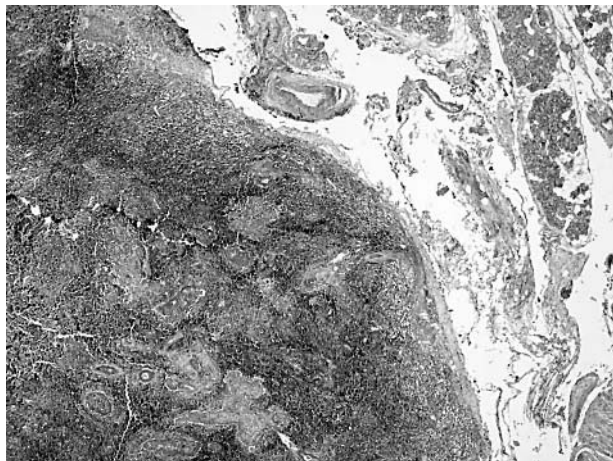
The patient is well 12 months after surgery.

### DISCUSSION

Lymphoid and epithelial lesions may arise from intraparotid LNs, which originate from the inclusions of salivary ducts and acini.<sup>2-7</sup> The frequency of these lesions increases dramatically when cystadenoma lymphomatousum cases, which are thought to arise from intranodal ductal inclusions, are considered.<sup>8</sup>

Multiple lumps at this region are frequently related to lesions originating from LNs, and the leading lesion is again cystadenoma lymphomatousum.<sup>9</sup> In our present case, histopathological examination revealed extensive SM of the

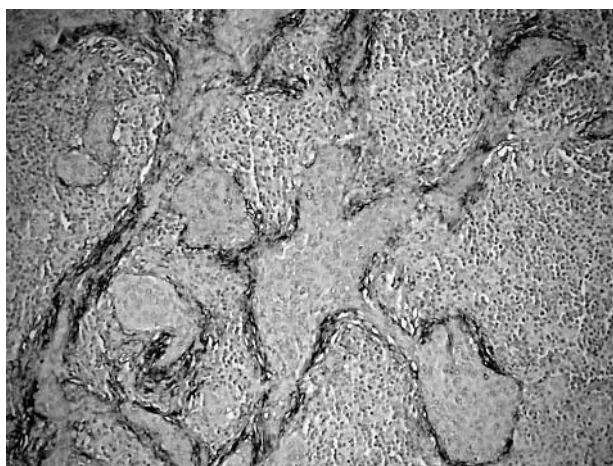
**Abbreviations:** LN, lymph node; PS, proliferative sialometaplasia; SM, squamous metaplasia



**Figure 2** Extensive squamous metaplasia of the intraparotid ductal inclusions; note the capsule of the lymph node and preserved architecture of the adjacent parotid gland (haematoxylin and eosin stain; original magnification,  $\times 4$ ).



**Figure 3** High power view of squamous metaplasia and nodal inclusions (haematoxylin and eosin stain; original magnification,  $\times 20$ ).



**Figure 4** Smooth muscle actin expression at the periphery of the metaplastic lobules (haematoxylin and eosin stain; original magnification,  $\times 10$ ).

### Take home messages

- We report a unique case of proliferative sialometaplasia of multiple intraparotid lymph nodes occurring in a 55 year old woman, presenting with multiple parotid lumps
- This interesting case points to the importance of intraparotid lymph nodes as sites for multiple lesions of the parotid region

intranodal inclusions, with a preserved lobular architecture involving five intraparotid LNs. Among the previous reports we noticed cases comparable to this one. Goldman and Klein<sup>2</sup> described PS of an intraparotid lymph node. Ryan *et al* described two patients with AIDS who presented with lumps at the parotid region, with lymphadenitis and SM of the salivary gland ductal inclusions.<sup>3</sup> As in our present case, those cases did not fulfill the established criteria for the diagnosis of necrotising sialometaplasia<sup>1</sup> described by Abrams *et al*,<sup>1</sup> because we could not consistently demonstrate infarction. Anneroth and Hansen<sup>10</sup> suggested five stages in the course of necrotising sialometaplasia, namely: infarction, sequestration, ulceration, reparation, and healing. The lesions in our patient might represent the reparation and healing stages of necrotising sialometaplasia, but without evidence of necrosis the case falls into the category of proliferative SM.

“We feel that this unique case is important because, to the best of our knowledge, this is the first case of proliferative sialometaplasia involving multiple intraparotid lymph nodes”

The differential diagnosis of proliferative SM also includes metastatic squamous cell carcinoma and mucoepidermoid carcinoma.<sup>2 10 11</sup> In our present case, no cellular atypia or mitotic activity was encountered, thus excluding carcinoma. The coexpression of cytokeratin, S-100, and actin was consistent with a lesion related to myoepithelial cells. The lack of chondromyxoid stroma and the presence of multiple intranodal lesions aided in the exclusion of squamous metaplasia prominent pleomorphic adenoma.<sup>2</sup> The patient's clinical features were inconsistent with the established criteria<sup>12</sup> of Sjögren's syndrome and the involvement of intraparotid lymph nodes has not been described for this disease.

We feel that this unique case is important because, to the best of our knowledge, this is the first case of PS involving multiple intraparotid LNs. PS should be included in the differential diagnosis of multiple lumps of the parotid region.

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## ECHO

## Familial frontotemporal dementia: early manifestations



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**F**rontotemporal dementia (FTD) is second only to Alzheimer's disease as a cause of dementia. It is mainly sporadic but may be familial, often with a mutation (P301L) in the *tau* gene. Behavioural and language disturbances are common and it may be associated with parkinsonism in some cases and motor neurone disease in others. The detailed findings in an asymptomatic woman with this mutation have been described from Italy.

Two of the subject's three brothers had developed FTD. Her one sister and the third brother were asymptomatic and neurologically and psychometrically normal at age 58 and 52 respectively and did not carry the mutation. The P301L mutation was demonstrated in the asymptomatic subject and her two affected brothers. At the age of 50 she was holding down a demanding job, leading an appropriate social life, and showed no behavioural abnormalities. She performed normally on a battery of cognitive tests with the single exception of the Verbal Fluency Test for letters in which she produced only 17 words in one minute compared with the 30 and 43 words of her two unaffected siblings. Brain CT and gross inspection of brain single photon emission computed tomography (SPECT) showed no abnormality but on SPECT with statistical parametric mapping (SPECT-SPM) there was reduced blood flow in the frontal lobes, particularly the dorsolateral frontal cortex and frontal poles and the mesial frontal cortex. SPECT-SM on the normal sister was within normal limits. The subject's cerebrospinal fluid (CSF) contained increased levels of A $\beta$ 1-42, tau protein, and 181P-tau. (Patients with Alzheimer's disease have high tau and low A $\beta$ 1-42 levels in CSF.)

Testing for the abnormalities found in this asymptomatic subject might improve early diagnostic accuracy in FTD and help to distinguish it from Alzheimer's disease.

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## Familial frontotemporal dementia: early manifestations

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