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Radiology in focus

A severe form of Sjogren's syndrome

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Abstract
Sjogren's syndrome is an autoimmune disease characterized by exocrine gland destruction and manifested by parotid, submandibular and lacrimal gland infection. We report a case with recurrent severe parotid gland infections. The sialographic and CT findings are presented.

Key words: Sjogren's syndrome: Diagnostic imaging

Case report

A 48-year-old woman presented with a bilateral, painful parotid gland swelling. During a period of 10 years, she had had several such episodes which had been treated by parenteral antibiotics. She also complained of xerostomia, xerophthalmia and rheumatoid arthritis. The patient had Sjogren's syndrome histologically diagnosed by an oral cavity minor salivary gland biopsy five years earlier.

Otolaryngological examination revealed both parotid glands to be enlarged and tender. The overlying skin was red and hot. Purulent secretion was observed at the orifices of Stensen's duct.

The patient was treated by intravenous flucloxacillin and metronidazole. She gradually improved over a period of 10 days.

A sialographic study revealed a normal main duct and multiple globular collections of contrast medium uniformly distributed throughout the gland. No calcifications were seen (Figure 1).

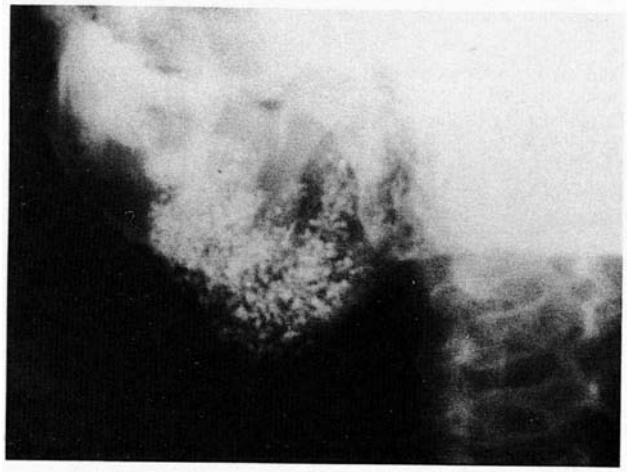

Fig. 1

Lateral oblique sialogram showing normal main duct and multiple globular collections of contrast medium, demonstrating pseudosialectatic changes.

Contrast enhanced transverse axial computed tomography (CT) showed bilateral parotid gland enlargement with an inhomogeneous appearance. The glands showed increased density, more evident on the right side. Numerous small low attenuation areas were noted. Scattered calcifications were seen on the right and punctate calcifications were noted inside a cystic area (Figure 2a, b).

Discussion

Sjogren's syndrome is an inflammatory autoimmune condition involving the salivary and lacrimal glands. It is characterized by a lymphocyte-mediated destructive process leading to xerostomia and keratoconjunctivitis sicca.

Sjogren's syndrome is now classified into two distinct entities: (i) primary involving the exocrine glands only; (2) secondary associated with a definable autoimmune disease, most commonly rheumatoid arthritis (March *et al.*, 1989). In the majority of cases this syndrome occurs in older women with an average onset age of 50 years.

The clinical picture is characterised by oral discomfort and a 'sandy' sensation in the eyes. Permanent or intermittent salivary gland swelling (usually parotid) occurs in more than 75 per cent of patients.

Diagnostic evaluation includes: conventional sialography (Som *et al.*, 1981; Iko, 1984; Takashima *et al.*, 1991); radionuclide scintigraphy (March *et al.*, 1989); ultrasound examination (Bradus *et al.*, 1988); computed tomography (Stone *et al.*, 1991; March *et al.*, 1989); and magnetic resonance imaging (Takashima *et al.*, 1991). However, biopsy of the salivary gland is mandatory for a definite diagnosis in order to exclude lymphatic degeneration.

Although CT and MRI have become the imaging modalities of choice in cases of parotid gland lesions, sialography is still a reasonable alternative especially in countries where newer imaging techniques are not readily available. We limited our investigation to conventional sialography and intravenous contrast enhanced CT examination which showed that characteristic changes of the disease: other radiological examinations were not necessary because of the obvious radiological picture obtained.

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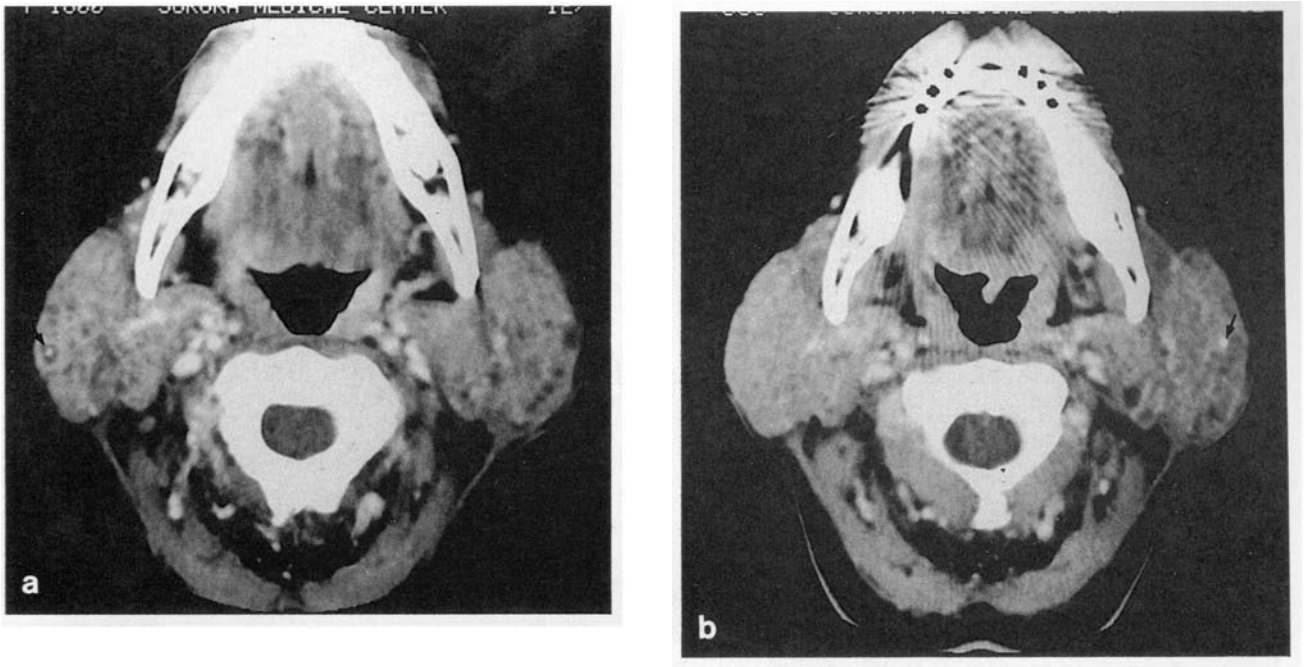

FIG. 2

(a) Intravenous contrast enhanced axial CT showing bilateral inhomogeneous parotid gland enlargement with numerous small cystic areas. A small calcification is seen in the dilated canal on the right hand side (arrowed); (b) A higher section of intravenous contrast enhanced axial CT showing a small calcification (arrowed) in the enlarged parotid gland.

Despite the fact that sialography is not a sophisticated procedure, it is a dependable modality for staging Sjogren's syndrome (Som *et al.*, 1981; Iko, 1984; Takashima *et al.*, 1991). Important staging features, well illustrated by sialography, include: normal or dilated ducts; punctate or globular collections; cavitary lesions; and destructive changes. Conventional sialography of our patient showed multiple globular collections of contrast medium measuring more than 2 mm in diameter and distributed uniformly throughout the gland. These features are consistent with the globular pseudoectatic stage of parotid inflammatory disease.

CT examination differentiates between intrinsic and extrinsic parotid masses. It also allows for evaluation of disease extension, including the location and possible involvement of the facial nerve. Local complications, including lymphoma, pseudolymphoma and abscess formation are also illustrated. Intravenous contrast enhancement aids in visualization of the vascular structures, distinction between intra- and extravascular calcification, better delineation of cystic areas and identification of lymphadenopathy (March *et al.*, 1989).

Most authors prefer CT–sialography, without contrast administration, in the evaluation of Sjogren's syndrome (Bradus *et al.*, 1988). In our case the presence of grossly dilated globular ducts made contrast injection into the Stensen's duct unnecessary. Scattered calcification throughout the gland represents microlithiasis and 4 mm diameter, cystic areas with punctate calcification are clearly seen.

We think that in the presence of characteristic conventional sialographic changes, the intravenous contrast CT examination (without contrast injection of Stensen's duct is sufficiently diagnostic.

Ultrasound examination, as a useful and rapid screening, is inexpensive and noninvasive but not sensitive nor specific. In most cases of non-complicated Sjogren's syndrome ultrasound shows hypoechoic solid masses (Bradus *et al.*, 1988; De Clerck *et al.*, 1988).

Magnetic resonance imaging (MRI) is an expensive examination. Depending on the stage of the disease normal signals or signals of inhomogeneous intensity are seen. Salt and pepper images are highly characteristic (Takashima *et al.* 1991).

Conclusion

Despite MRI being the imaging modality of choice for parotid gland pathology, we think that conventional sialography and intravenous enhanced CT are good alternatives for the diagnosis of Sjogren's syndrome with multicystic degenerative changes.

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