CASE REPORT

Intraparotid ductal ectasia: rare cause of parotid swelling

Houda Chahed, Samia Meherzi, Azza Mediouni, Mohamed Ben Amor

Department of ENT, Hôpital la Rabta, Tunis, Tunisia

Correspondence to

Dr Houda Chahed,
houda.chahed@gmail.com

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SUMMARY

A 41-year-old patient was hospitalised for a chronic right parotid mass. A cervical ultrasound revealed a cystic mass of the parotid. Cervical MRI found a ductal ectasia of the parotid and submandibular glands associated with a retention cyst of the right parotid. He had a right total parotidectomy. Histopathological examination of the lesion revealed a multilocular cystic mass with a diffuse glandular ectasia of salivary ducts. The patient had an uneventful postoperative course without any recurrence of symptoms.

BACKGROUND

Sialectasis is an uncommon condition characterised by abnormal dilation of salivary ducts. It can result from several causes including sialoliths, recurrent infection, trauma or congenital abnormalities.¹ Diffuse dilatation of intraglandular ductal system is an extremely rare disease.^{1,2} Most often, this condition affects the parotid gland and largely spares the submandibular gland.² To the best of our knowledge, there are no reports about idiopathic intraglandular ductal system ectasia of both parotid and submandibular glands with large unilateral sialectasis of the Stensen's duct. In this report, we describe the clinical and radiographic features of our case report and discuss therapeutic management.

CASE PRESENTATION

A 41-year-old man presented with a 17-year history of painless, slowly progressive swelling of the right parotid region. The patient had no history of parotid sialadenitis, facial trauma or allergic disease. There was no increase in size related to eating. Physical examination revealed a fluctuant, 7 cm swelling of the right parotid gland (figure 1). The overlying skin had a normal appearance. The Stensen's duct ostium was normal. No obstruction to saliva flow was identified. The other major salivary glands were clinically asymptomatic. No lymph node abnormality was observed.

INVESTIGATIONS

On ultrasound imaging, a well-defined cystic mass was noted. There was no evidence of calculi or abscess. On MRI, both T1-weighted and T2-weighted images showed diffuse ectasia of salivary ducts within both parotid and submandibular glands. The extent of enlargement was greater in the right parotid gland with a large well-delineated

and homogeneous cystic lesion following the route of the Stensen's duct, not enhanced after the injection of gadolinium (figures 2 and 3). Fine-needle aspiration cytology showed the presence of inflammatory fluid.

Laboratory results didn't show hyper leucocyte or hyper eosinophily. HIV serology was negative.

DIFFERENTIAL DIAGNOSIS

Based on the radiologic findings, Sjogren's syndrome was suspected but a biopsy of minor salivary glands of lower labial mucosa and Shirmer's test were negative. Other differential diagnoses were suspected such as sialodochitis fibrinosa, sialocoele, sialolithiasis, ductal stenosis and polycystic disease. They were ruled out based on the uneventful patient history, radiologic findings and laboratory test results.

TREATMENT

Repeated aspiration was attempted but was associated with early recurrence. Hence, total parotidectomy was performed with preservation of the facial nerve. Histopathological examination of the lesion revealed a multilocular cystic mass with glandular ectasia of salivary ducts.

OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative course without any recurrence of symptoms. Facial nerve function was normal. No recurrence on others salivary glands was described during 4 years of follow-up.

DISCUSSION

Constitutional sialectasis is one type of rare heteroplasia of the salivary glands, which can involve the ducts of the parotid and submandibular glands.¹ Seifert *et al* reported only 5 cases of congenital sialectasis among the 360 cases of non-neoplastic cystic lesions of the salivary glands.³ This hypothesis was the most accepted in our case given multifocal ductal ectasia of major salivary glands without any obvious cause of obstruction. Search for an autoimmune disease that may explain this symptom such as Sjogren's syndrome or sarcoidosis was negative in our case.

Clinically, it presents as a painless swelling in the cheek that is not related to eating and may be unilateral or bilateral.⁴

Sialography, CT scan and MRI are considered to be the most accurate investigations in the diagnosis

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Figure 1 Pretherapeutic aspect of the parotid swelling.

of dilatation of salivary ducts.^{5 6} Sialography shows dilatation of the salivary ducts with a well-defined border. CT scan can demonstrate the presence or the absence of a sialolith. MRI is

Figure 2 Parotid MRI with axial T1-weighted image showing a homogeneous cystic lesion of the superficial lobe of the right parotid gland, hypointense on the T1-weighted image (red arrow).

Figure 3 Parotid MRI with coronal T2-weighted image showing a homogeneous cystic lesion of the superficial lobe of the right parotid gland, hyperintense on the T2-weighted image (red star) with ectasia of salivary ducts in bilateral parotid (yellow arrow) and submandibular glands (red arrow).

the most effective method demonstrating location and degree of stricture or dilatation. MRI is the gold standard for exploring salivary glands.^{4 5} Indeed, MRI and sialo-MRI showed ductal ectasia with salivary retention and cystic mass.³

Differential diagnoses of this exceptional pathology arises with other disorders of salivary glands such as polycystic parotid, ductal stenosis and also with systemic sialosis mainly Sjogren's syndrome, sarcoidosis and Kimura disease.^{6 7}

Polycystic disease is a rare disorder of the ductal system of the parotid salivary glands. A female predilection has been reported. Clinically, it is characterised by recurrent painless swelling of the involved gland. Histopathologically, cysts of varying size are found to replace the gland parenchyma.⁸

An autoimmune disease such as Sjogren disease might be considered. However, such patients might have decreased salivary flow and associated systemic findings.⁷ It was negative in our case.

Sialodochitis fibrinosa is another rare differential diagnosis. But our patient had no history of allergy. No abundant eosinophils were found in the haemogram. Imaging studies, such as MRI and sialography, demonstrate dilated salivary ducts and ectasia. Salivary gland histology did not show lymphocytic infiltration with abundant eosinophils.⁹

Several treatment options are available for this condition. The choice of therapeutic modality depends on the size, the location of dilatation and the surgeon's experience.

Conservative methods like repeated aspiration and compression, dilation of the papilla and stent placement may be performed but usually associated with recurrence.² In such case, a more aggressive surgical management may be indicated such as marsupialisation for distal sialectasis.² Currently, advances in minimally invasive surgery and interventional endoscopy have reduced the use of aggressive surgical treatment.^{10–12} Nahlieli *et al* reported

a success rate of 81% for the treatment of stenosis by sialendoscopy.⁹

In others reports, more aggressive surgical management has been indicated for distal dilatation of salivary duct, such as excision of the dilated portion of the duct, reimplantation into buccal mucosa and superficial parotidectomy and excision of the Stensen's duct to be an effective method of treatment.^{2 12} In our case, repeated aspiration was associated with early recurrence. We have no experience with sialendoscopy and dilatation. Ectasia of intraparotid ducts was diffuse affecting both terminal and proximal ducts and causing an important enlargement of the gland and a considerable discomfort for the patient. Consequently, we performed a right total parotidectomy as superficial one may be complicated with sialocoele formation.²

Learning points

- ▶ Intraglandular ductal ectasia is an exceptional entity. Its diagnosis is guided by imaging mainly sialo-MRI.
- ▶ Aetiological investigation is needed to rule out systemic disease.
- ▶ Interventional endoscopy have reduced the use of aggressive surgical treatment but parotidectomy remains an effective treatment of this pathology.
- ▶ A long-term periodic review is essential to evaluate ductal ectasia in others salivary glands.

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