

Scintigraphic detection of a parotid salivary gland malfunction, in chronic sialolithiasis and fat infiltration with no risk factors

Iordanis Konstantinidis¹ MD, PhD,
Evangelia Tsakiropoulou¹ MD,
Angelos Chatziavramidis¹ MD,
Ioannis Iakovou² MD, PhD

1. 2nd Academic ORL Department
and

2. Academic Department
of Nuclear Medicine,
Aristotle University of Thessa-
loniki, Thessaloniki, Greece

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Correspondence address:

Dr. Iordanis Konstantinidis
3A Kapetan Kotta str,
P. C. 55236 Panorama,
Thessaloniki, Greece
Email: jordan_orl@hotmail.com
Tel: 00302310321677
Fax: 00302310934888

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Abstract

Long lasting sialolithiasis can cause functional and structural changes of the salivary glands, inflammatory infiltration and fibrosis. However fat infiltration with sialolithiasis has not been described in the parotid glands. We describe a 60 years old man, bus-driver who presented with a history of recurrent right parotid sialadenitis and was diagnosed to have bilateral sialolithiasis and left parotid fat infiltration. Imaging showed large intraparenchymal stones in both parotid glands. Gland atrophy with homogeneous fat distribution and severe hypofunction were the main imaging findings on the left side. The right parotid gland had normal findings in imaging studies. In conclusion, we suggest that sialolithiasis caused chronic obstruction, due to increased ductal pressure, sialadenitis, fat infiltration, hypofunction and atrophy of the left parotid gland. Patient denied further treatment.

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Introduction

Sialolithiasis of the salivary glands can cause chronic obstruction resulting into permanent parenchymal changes like inflammatory infiltration and fibrosis [1]. Fat infiltration of the parotid gland is often observed in elderly people as well as, in Sjogren's syndrome, sialadenitis and in some diseases related to fat metabolism [2-4]. However, bilateral parotid sialolithiasis with fat infiltration in one parotid gland and severe atrophy of this gland have not been previously scintigraphically described.

Case presentation

A 60 years old man, bus-driver presented with a 4 years history of recurrent right parotid painful but feverless swellings related with meals, sometimes lasting for more than 2 days. The last such episode was 3 weeks ago. At the left parotid, 8 years ago, he had 3 such episodes within one year. These episodes were treated conservatively with gland massage and when symptoms lasted more than two days, antibiotics were also administered. The patient had no xerostomia and no difficulty in mastication, or swallowing. Furthermore, no history of alcoholism, thyroid disorders, anorexia, bulimia, diabetes mellitus, hyperlipidemia or any autoimmune disease had been reported. Clinical examination did not reveal swelling, palpable mass or significant enlargement of the parotids. Bimanual palpation of the glands induced normal salivary flow from the right but not from the left Stensen's duct.

Computed tomography (CT) scan of the head demonstrated an intraparenchymal stone in each of the parotid glands. The right sialolith was 0.8x0.5cm in size with normal parotid parenchyma (Fig. 1). The left sialolith was 1.2x0.5cm. Both sialoliths were located anteriorly within the parenchyma just posterior to the masseter muscle. The left parotid gland additionally showed fatty appearance and diffuse atrophy (Fig. 1). Further imaging with magnetic resonance imaging (MRI) showed a homogeneous adipose tissue deposition within the parenchyma. Specifically, left parotid gland in T2 weighted MRI sequences had a pathognomonic hypersignal for fat deposition (Fig. 2). The left parotid gland was atrophic, only 2/3 of the size of the right one, without cystic degeneration (Fig. 2). Fine needle aspiration cytology examination was inconclusive showing atrophic glandular cells with sparse adipose tissue distribution, mixed with blood, inadequate for diagnosis. An open biopsy was avoided since it was considered as not necessary.

Scintigraphy of the salivary glands was also performed in order to assess their functional. Dynamic imaging of the whole anterior part of patient's head started after a bolus

of intravenous injection of 185MBq ^{99m}Tc -pertechnetate, at 1 frame per 30sec for 30min. At 15min after injection, diluted lemon juice was orally administered. Two functional parameters were described after analysis of the dynamic study time-activity curves: uptake rate, taken as the value of the initial slope of the time-activity curve, and washout fraction, which was the relative mobilizable radioactivity from each parotid gland after ingestion of the sialogogue. Severe dysfunction of the left parotid gland was demonstrated, with mean uptake values for the right and left gland of 0.34% and 0.09%, respectively (normal range 0.6%-0.17%) (Fig. 3). The washout fraction of the tracer after lemon juice stimulation was 60% for the right and 5% for the left parotid gland (normal range 61%-48%). Submandibular glands function was normal, with uptake values for the right and left gland: 0.35% and 0.3%, and washout fractions of 39% and 40% (normal range 0.5%-0.15% and 50%-20%, respectively).

Blood examinations for liver function were normal and for autoimmune disease were negative. The right sialolith was removed by sialendoscopy and transcutaneous approach under general anesthesia. No further treatment for the left side was decided due to the absence of symptoms and to patient's preference. The patient remained free of symptoms during one year of follow-up.

Discussion

Fat infiltration of the major salivary glands has been reported in several conditions: as a normal aging process in sialadenosis, in diseases related to fat metabolism, as in alcoholism, malnutrition, anorexia, bulimia, hypothyroidism, diabetes mellitus, in HIV infections and in autoimmune disorders like Sjögren's syndrome [2-4]. Furthermore, gland atrophy and fat infiltration can occur after radiation exposure [5]. To our knowledge, fat infiltration with sialolithiasis has

never been reported in a parotid gland. This is in contrast with the existed literature for the submandibular gland where sialolithiasis and fat deposition have a well recognized relation [6, 7]. Previous medical reports regard to normal saliva flow in the past do not suggest a congenital hypofunction. In addition, although aging could be a cause of fat deposition, this is not in accordance with unilateral findings.

Fat deposition usually results in adipose tissue infiltration and/or acinar cells and whole gland hypertrophy. The parotid function usually remains intact as in sialadenosis [3]. According to Carda et al (2005), the histological pattern of fat infiltration differs between diabetics and alcoholics [4]. Abundant adipose infiltration in the stroma and numerous lipid intracytoplasmic droplets in the acinar and ductal cells have been observed in diabetics. Fat deposition in Sjögren syndrome is diffusely scattered, but this pattern doesn't seem to be disease specific [8]. On the contrary, in cases of chronic parotitis, fat is localized in nodule-like areas and not as in our case, diffusely scattered. Other researchers have also described differences in fat deposition of submandibular salivary glands between aging and chronic inflammation [9]. In older people, fat is prominent in the secretory and duct cells, while in inflammation is found in the histiocytes located near small abscesses.

In our case the exact mechanism of fat infiltration was not clear. It is uncertain whether chronic inflammation can trigger a progressive replacement of normal parenchyma by adipose tissue as no findings of chronic inflammation, like cystic degeneration or enlarged intraparenchymal lymph nodes were found in the imaging studies. Acute obstructive episodes causing increased intraductal pressure and accumulation of saliva may result in hypoxia of the parotid parenchyma stimulating the precursor cells to become adipose cells. Indeed, in a case of a submandibular fat infiltration mentioned above [7], the authors suggested that increased pressure of the ductal system due to long-term

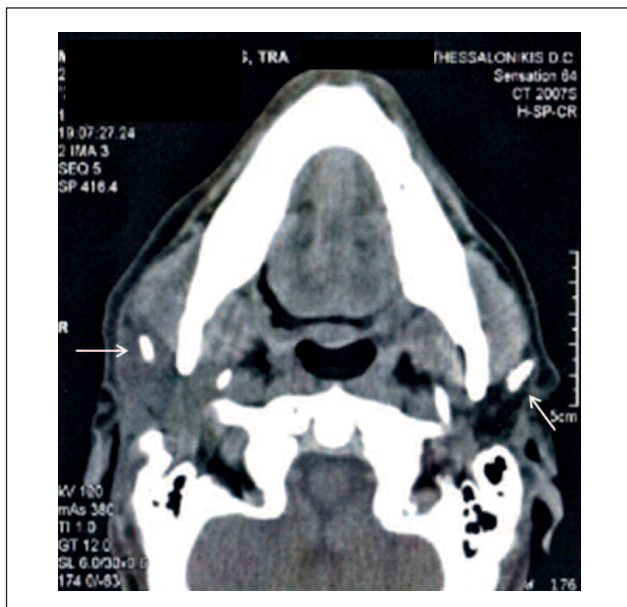


Figure 1. The CT scan of the head showed intraparenchymal stones in both sides (white arrows) and the dark fatty appearance of the left parotid gland. The right parotid gland had normal parenchymal appearance.

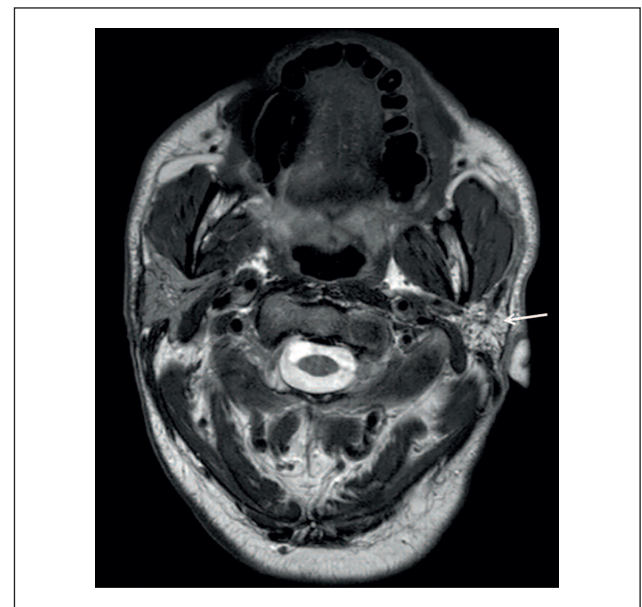


Figure 2. The MRI examination with T2 weighted sequence TSE showed a homogeneous fat infiltration (hypersignal) of the left parotid gland (white arrow) along with a significant parenchymal atrophy.

stasis led to anoxia of the gland, which caused acinar cells atrophy and replacement by fat tissue [7].

Experimental studies in rats showed that parotid duct ligation for one week caused atrophy of the parotid glands [10]. In another study in rats, long term duct obstruction enhanced apoptosis of duct and capillary endothelial cells sug-

gesting that distorted capillary blood supply was related to gland atrophy [11]. This may explain atrophy of the parenchyma but not adipose deposition. It has also been reported that removing obstruction resulted in regeneration of acinar cells from precursor cells in the residual ducts [11]. It seems that an experimental study is difficult to perform due to the long process needed for fat deposition in human salivary glands.

In conclusion, based on the history of the presented case it seems that repeated acute episodes of sialadenitis are more likely to induce anoxia conditions to the parotid parenchyma rather than chronic inflammation. This may cause a “quick death” of the acinar cells, being the triggering factor for a long replacement process by adipose tissue. Further studies are needed to explain the pathogenesis and its clinical significance.

The authors declare that they have no conflicts of interest.

Bibliography

- Harrison JD. Causes, natural history, and incidence of salivary stones and obstructions. *Otolaryngol Clin North Am* 2009; 42(6): 927-47.
- Garcia DS, Bussoloti Filho I. Fat deposition of parotid glands. *Braz J Otorhinolaryngol* 2013; 79(2): 173-6.
- Guggenheimer J, Close JM, Eghtesad B. Sialadenitis in Patients with Advanced Liver Disease. *Head and Neck Pathol* 2009; 3: 100-5.
- Carda C, Carranza M, Arriaga A et al. Structural differences between alcoholic and diabetic parotid sialosis. *Med Oral Patol/Oral Cir Bucal* 2005; 10: 309-14.
- Harrington AC, Stasko T. Radiation-induced adiposis of the parotid gland. *J Dermatol Surg Oncol* 1994; 20(4): 246-50.
- Sumi M, Izumi M, Yonetsu K, Nakamura T. The MR imaging assessment of submandibular gland sialoadenitis secondary to sialolithiasis: correlation with CT and histopathologic findings. *Am J Neuro-radiol* 1999; 20(9): 1737-43.
- Hiramoto R, Ishibashi K, Asada K et al. Case of sialolithiasis with severe fatty change of the submandibular salivary gland. *Tsurumi Shi gaku* 1989; 15(3): 427-31.
- Izumi M, Eguchi K, Nakamura H et al. Premature fat deposition in the salivary glands associated with Sjögren syndrome: MR and CT evidence. *Am J Neuroradiol* 1997; 18(5): 951-8.
- Garrett JR. Some observations on human submandibular salivary glands. *Proc Royal Soc Med* 1962; 55: 488-91.
- Burgess KL, Dardick I. Cell population changes during atrophy and regeneration of rat parotid gland. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998; 85(6): 699-706.
- Walker NI, Gobé GC. Cell death and cell proliferation during atrophy of the rat parotid gland induced by duct obstruction. *J Pathol* 1987; 153(4): 333-44.

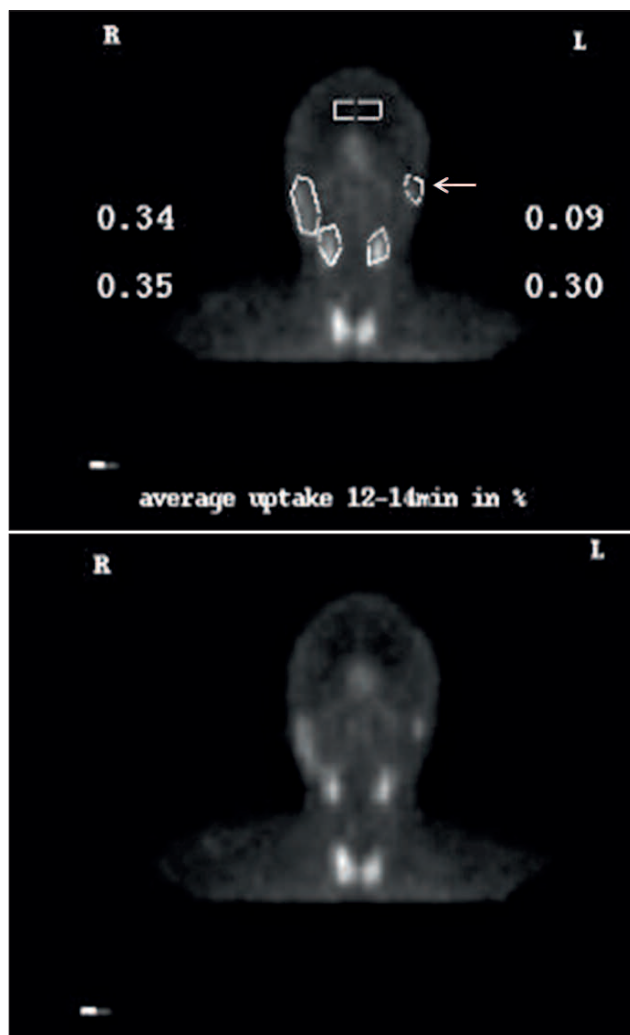


Figure 3. Salivary glands scan with $^{99m}\text{TcO}_4$ showed severe hypofunction of the left parotid gland (arrow) with very low uptake of the tracer. The first line of numbers indicates the tracer's uptake of the parotids; rate (%) at 20min and the second line, the uptake of the submandibular glands.