J Oral Pathol Med (2004) 33: 441–2

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CASE REPORT

Anorexia/bulimia-related sialadenosis of palatal minor salivary glands

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In patients affected by alimentary disorders sialadenosis is frequently observed. This non-inflammatory condition is described to affect major salivary glands, leading to the characteristic parotid and/or submandibular swelling. Thus fine-needle aspiration cytology or parotid open biopsy are generally required to diagnose histologically the disorder. We report the case of a 28-year-old patient affected by bulimia/anorexia nervosa who presented, in addition to parotid enlargement, a bilateral symmetric painless soft swelling of the hard palate. The lesion was biopsed and histopathological examination showed the classical features of sialadenosis. To our knowledge, this is the first case of sialadenosis affecting palatal minor salivary glands. It underlines that when sialadenosis is clinically suspected, clinicians could check also patients' oral cavity for minor salivary glands involvement, in order to potentially avoid invasive extra-oral procedures and to easily confirm diagnosis with an intra-oral biopsy.
J Oral Pathol Med (2004) **33**: 441–2

Keywords: anorexia; bulimia; minor salivary glands; sialadenosis

The patient was a 28-year-old woman who was diagnosed to be affected by eating disorders since 1998 and referred to our Division of Oral Medicine for evaluation of persistent parotid swelling. On extra-oral examination a bilateral symmetrical enlargement of the parotid glands was evident (Fig. 1). The swelling was painless and non-tender to palpation, and no cervical lymphadenopathy was present. Intra-orally the parotid duct orifices appeared normal and no duct obstructions were evident; milking of the parotids produced a normal salivary flow.

A more careful intra-oral examination revealed the presence of a bilateral symmetrical swelling of the hard palate which was covered by normal mucosa and showed to be painless and soft to palpation (Fig. 2).

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Accepted for publication October 29, 2003

On the basis of the patient's medical history, a clinical diagnosis of bulimia/anorexia-related sialadenosis of the parotids and minor palatal salivary glands was suggested.

A computed tomography (CT) scan of the parotids showed a wide bilateral enlargement of the parotid glandular parenchyma containing many small calcifications. Further diagnostic evaluation included an incisional biopsy of the palatal swelling and laboratory studies aimed at evaluating other possible causes of chronic asymptomatic salivary glands swelling. Thus, antinuclear antibody (ANA), antibodies to SS antigen A (SS-A/Ro), SS antigen B (SS-B/La), and hepatitis C virus (HCV), rheumatoid factor, full blood count, serum angiotensin-converting enzyme (ACE), albumin, immunoglobulin (Ig)A, IgG, IgM, glutamic oxaloacetic transaminase (GOT), glutamic pyruvic transaminase (GPT), fasting blood glucose, and urine Bence-Jones protein were evaluated and resulted into normal limits.

Histopathological examination of the specimen revealed hypertrophy of the acinar cells with small, round, and basally situated nuclei, together with areas of fatty infiltration. No inflammatory cells were visible.

Thus, the patient was diagnosed to be affected by bulimia/anorexia-related sialadenosis of the major and minor salivary glands.

With regard to therapy, in addition to non-treatment and surgical approaches, pilocarpine has been recently showed to be effective in reducing parotid glands enlargement in bulimic patients (1). Accordingly, we administered to our patient pilocarpine hydrochloride drops orally at the dosage of 2.5 mg bid., and we obtained a relevant reduction of parotid and palatal swellings over a period of 2 months, without significant side effects.

Comments

Sialadenosis refers to non-inflammatory enlargement of the salivary glands which is almost always associated with an underlying systemic disorder, such as diabetes, alcoholism, malnutrition, anorexia nervosa, bulimia and others (2, 3). It is thought that the various causes

Figure 1 Bilateral symmetrical enlargement of the parotid glands.

Figure 2 Bilateral symmetrical swelling of the hard palate. The lesions were covered by normal mucosa and showed to be painless and soft to palpation.

of sialadenosis all result in a common pathogenetic effect in that they produce a peripheral autonomic neuropathy which is responsible for disordered metabolism and secretion, resulting in acinar enlargement (2).

With regard to anorexia/bulimia-related sialadenosis, it is reported to develop in 10–60% of the patients affected by these complex psychological eating disorders (2), and in some cases it has been described as their presenting sign (2, 4).

Major salivary glands, mainly parotids, are usually affected, leading to the characteristic parotid and/or submandibular swelling.

Thus fine-needle aspiration cytology (FNAC) is generally performed to confirm the diagnosis (2, 5). However, FNAC requires the assistance of an expert pathologist and has his main value in the exclusion of a tumour and the assessment of acinar size (Fig. 3) (5).

So, a parotid open biopsy may be necessary to confirm the diagnosis of sialadenosis (2, 5).

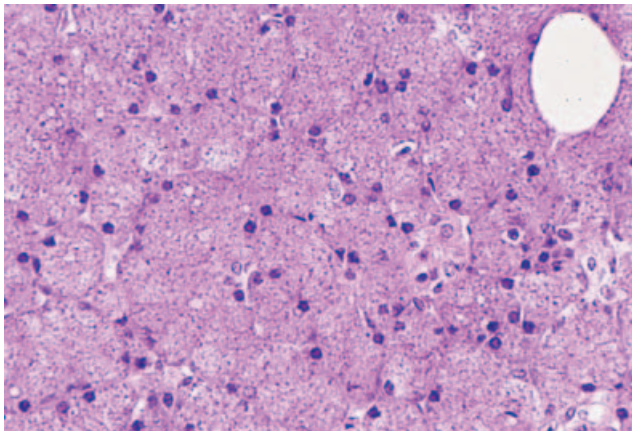

Figure 3 Hypertrophy of the acinar cells with small, round, and basally situated nuclei. Fatty infiltration without inflammatory cells is visible (2–5).

Nevertheless, performing facial surgery in patient affected by eating disorders as consequence of altered self-esteem and self-perception may represent a serious risk. Accordingly, some authors believe that surgical management of parotid enlargement in patients with bulimia nervosa is contraindicated (6).

From this point of view our case is interesting as the histological diagnosis was made by performing easily an intra-oral biopsy of the palatal minor salivary glands which clinically appeared enlarged and involved by the disorder.

To our knowledge, this is the first case of minor salivary glands involvement in a patient affected by anorexia/bulimia-related sialadenosis. Even if further and larger studies are clearly needed, this report underlines that for minor salivary glands involvement could be useful in order to avoid potentially invasive extra-oral procedures and to easily confirm sialadenosis diagnosis with an intra-oral biopsy.

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