Bilateral Parotid Sialadenosis Associated with Long-Standing Bulimia: A Case Report and Literature Review

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Abstract

Purpose Bulimia is a common cause of sialadenosis. This paper presents a case of bilateral parotid sialadenosis associated with long-standing bulimia, and reviews the relevant literature and current treatment options.

Methods and Results A 32-year-old woman had severe bilateral parotid sialomegaly for the last 6 years, which had occurred secondary to bulimia nervosa, which she had since 14 years. Treatment with pilocarpine was unsuccessful, so she underwent bilateral conservative parotidectomy. This procedure not only improved the aesthetic appearance of the patient but also improved her social and work life and overall quality of life.

Conclusions Sialomegaly secondary to bulimia results in a major alteration of the aesthetics of a patient's face. Conservative measures are not enough in many cases, and parotidectomy may be the only viable option, as it can also significantly improve adherence to psychiatric treatment for bulimia, in addition to correcting the facial aesthetics.

Keywords Bulimia · Parotid gland · Sialadenosis · Parotidectomy · Eating disorder

Introduction

Sialadenosis refers to enlargement of the salivary glands, and is non-inflammatory and non-cancerous in nature. Sialadenosis is typically a systemic disorder that is associated with the endocrine–metabolic system (in diabetes mellitus, dysfunction of gonads, pituitary gland, thyroid gland etc.); dystrophic–metabolic sialadenosis (malnutrition, avitaminosis, alcoholism, chronic liver diseases etc.) and neurogenic sialadenosis (dysfunction of the vegetative nervous system, drugs, antihypertensive agents, etc.) [1]. There are reports of bilateral parotid growth in individuals with high intake of foods rich in carbohydrates in underdeveloped countries, especially after the introduction of a normal diet after long periods of famine and extreme malnutrition [2]. However, in the Western world, the leading cause of bilateral parotid growth of an endocrine–metabolic non-inflammatory nature is eating disorders. Bulimia or bulimia nervosa is an eating disorder characterized by frequent bingeing and purging that is associated with many negative health consequences [3]. Lavender [4] first described the association between continuous vomiting and bilateral parotid growth in 1969, and the fact that bilateral parotid growth is sometimes the first visible manifestation of bulimia. Bulimia is a serious health problem in developed countries, with a prevalence of 1 % that is predominant in young women.

This paper presents one of 12 case reports of bilateral parotid sialadenosis associated with long-standing bulimia in the published literature to date and the most extensive review of the relevant literature and current treatment options for these patients.

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Case Report

We present the case of a 32-year-old woman with a history of bulimia nervosa for 14 years who had bilateral parotid gland enlargement since 6 years. The patient reported taking 600 mg ibuprofen every 12 or 24 h daily for 2–3 years to decrease the parotid swelling, and reported having gastric problems in the last few months. Physical examination showed bilateral parotid swelling that

significantly altered the aesthetics of the patient's face (Fig. 1). Palpation of both parotid glands did not reveal any tumor, as both glands were of a soft consistency, had not adhered to the deep planes, were not painful on palpation, and had preserved drainage salivary glands. A blood count test was performed, and the levels of serum angiotensin-converting enzyme (ACE), anti-Ro/Anti-LA antibodies, and c-ANCA/p-ANCA were determined. Analysis of these parameters showed no alteration in their normal values. Fine needle aspiration (FNA) revealed hypertrophy of acinar cells which supports a nonspecific origin of the sialadenosis. CT revealed considerable enlargement of both parotid glands (Fig. 2). The anteroposterior axis of the left parotid gland was of 8.6 cm; mediolateral axis, 6.1 cm; and craniocaudal axis, 6.9 cm. In the right parotid gland, the anteroposterior axis was 8.1 cm; mediolateral axis, 6.6 cm; and craniocaudal axis, 7.1 cm. No glandular masses or stones were found in the glandular parenchyma. Orthopantomography revealed multiple dental restorations because of several dental caries and multiple lost teeth rehabilitated by implantation (Fig. 3).

After ruling out other diseases, such as Sjögren's syndrome, alcoholism, and sarcoidosis that presented with non-inflammatory swelling of the salivary glands, sialadenosis secondary to bulimia nervosa was diagnosed.

Before treatment was started, we obtained the report of the patient's psychiatrist, who stated that her eating disorder had resolved 2 years ago. The psychiatrist added that

Fig. 1 Appearance of the patient before surgery. Images show bilateral parotid swelling that significantly alters the aesthetics of the patient's face

Fig. 2 TC in 3 plane with 3D reconstructions by Brainlab iPlan software 3.0.5, where considerable enlargement of both parotid glands can be seen. Both glandular lobes, superficial and deep (marked in red), present large hypertrophy

Fig. 3 Orthopantomography where several dental caries and multiple lost teeth rehabilitated by implantation were observed

Fig. 4 Appearance of the patient after surgery. Spectacular improvement of her cosmetic appearance can be seen

the bilateral sialomegaly negatively affected the self-esteem of the patient because it had significantly altered her appearance and therefore affected her social life; her psychiatrist also recommended that her sialomegaly be treated. Conservative treatment was initiated with oral pilocarpine administered at a dose of 15 mg/d (divided into three doses per day of 5 mg each) for 3 months, but the treatment was not successful. The patient then underwent surgical treatment with bilateral superficial parotidectomy. The large size of both the glands was rather unexpected, with the right parotid gland weighing 57 g and the left parotid gland weighing 62 g. Histopathological analysis revealed that both glands had large acinar cells that were three times the size of normal acinar cells (150 microns); moreover, the nuclei of the cells were displaced and the cells were filled with zymogen granules that were strongly positive for PAS, with mild lymphocytic infiltration and fat vacuoles. All these findings are indicative of granular-type sialadenosis.

After surgery, the patient reported improvement in her aesthetic appearance and quality of life, both her social and work life, and at 12 months after the procedure, the sialomegaly had not recurred (Fig. 4A, B).

Discussion

Bulimia has a prevalence of 1 % in the general population and up to 13 % in adolescents between 14 and 24 years; it is predominant in women aged 20 ± 4.3 years [5]. Sialomegaly secondary to bulimia occurs in 10–68 % of bulimic patients and is correlated directly with the number of episodes of self-induced vomiting in a day: it is typically found in patients who report at least 1–3 episodes of self-induced vomiting in a day [6, 7].

The pathogenesis of bulimic sialomegaly is not clear. It is speculated that sialomegaly is caused by repeated cholinergic stimulation of the parotid gland resulting from stimulation of the taste receptors on the tongue that occurs during vomiting. Such stimulation over a long period of time results in glandular hypertrophy. The trophic stimuli are produced by contact of proteolytic enzymes from the pancreas with the oral mucosa during vomiting [6–8]. Another theory is that the cholinergic stimulation that occurs during vomiting produces a significant increase in salivary flow with time, which leads to acinar hypertrophy and eventually parotid sialomegaly [8, 9].

The differential diagnosis includes all the disorders that are characterized by bilateral enlargement of the parotid glands (Table 1) [2, 10–12].

In cases of sialadenosis secondary to bulimia, surgeons should conduct a thorough medical history, which should include information about the usual diet of the patient, the type of physical activity the patient engages in, BMI, drugs commonly used, dental status (as bulimia is associated with an increase of caries, gum disease, and tooth loss), and psychiatrist evaluation. In addition, tests for blood count, biochemistry, and antibodies should be performed. Hypokalemia (serum K, <3.0 mmol/L) as a result of diuretics and laxatives is a common finding. Some of the other findings are changes in ECG reports, hypertriglyceridemia and hypercholesterolemia, increased lactate dehydrogenase (LDH), hyperuricemia, hypercalcemia, hyperglycemia, and increase in serum amylase (which is related to binge eating more than self-induced vomiting) [13, 14]. The levels of anti-Ro/anti-La, p-ANCA and c-ANCA, and ACE should also be determined in order to rule out Sjögren's syndrome, Wegener's granulomatosis, and sarcoidosis respectively, which are also characterized by enlargement of the parotid glands. FNA plays an important role in the diagnosis of sialadenosis, as this condition is characterized by an increase in the size of acinar cells as a result of increase in the secretion of enzyme granules. In addition, CT and MRI are crucial in the differential diagnosis, to rule out tumor, stones and intraparotid nodes, as the typical findings of sialadenosis are enlargement of both parotid glands, glandular parenchyma with increased granular density and

Table 1 List of differential diagnoses for bulimic sialomegaly

Congenital anomalies
<i>Trauma</i>
Infection
Viral (Paramyxovirus, Coxsackie virus B1/B6, HIV)
Pyogenic (mixed flora, <i>Streptococcus pyogenes</i> , <i>Staphylococcus aureus</i> , <i>Escherichia coli</i> , <i>Pseudomonas aeruginosa</i> , <i>Mycobacterium tuberculosis</i> ...)
Parasites: Chagas' disease
Fungal
Metabolic
Hypothalamus–hypophysis–adrenal axis disorder
Diabetes
Hypoparathyroidism
Hypercholesterolemia
Hypertryglyceridemia
Gout
Nutritional
Extreme malnutrition
Excessive starch intake
Vitamin deficiencies
Alcoholic cirrhosis
Obesity
Pseudo-parotomegaly
Masseter hypertrophy
Atrophy of other soft tissues with age
Nutritional
Extreme malnutrition
Excessive starch intake
Vitamin deficiencies
Pellagra and beriberi
Alcoholic cirrhosis
Autoimmune
Systemic lupus erythematosus
Sjögren's syndrome
Mikulicz's syndrome
Rheumatoid arthritis
Granulomatous disorders
Sarcoidosis
Wegener's granulomatosis
Histiocytosis X
Lymphoproliferative syndromes
Bilateral lymphoma
Castleman's disease
Drug induced (thiouracil, isoprenaline, dextropropoxyphene, 1st generation contraceptive pill, cytosine arabinoside, ritodrine, rinograffin, iodide, sulfisoxazole, phenylbutazone, phenytoin)
Allergic reactions
Toxicity
Iodine
Heavy metals

Table 1 continued

Congenital anomalies
Gastrointestinal diseases
Chronic pancreatitis
Zollinger–Ellison
Neurogenic
Autonomic dysfunction
Psychogenic (eating disorders)
Parotid cyst
Neoplastic
Parotid: benign and malignant tumors
Lymph node parotid metastases of cutaneous squamous cell carcinoma and melanoma of the head
Esophageal carcinoma
Unknown
Sialodochitis fibrinosa
Kussmaul's disease
Pneumoparotiditis
Testicular and ovarian atrophy

[2, 10–12]

variable fatty infiltration that could be misconstrued as a lipoma [11].

Sialography can be a useful tool in the diagnosis of sialadenosis especially in the later stages of the disease, when changes are caused by acinar swelling along with compression of the proximal ducts. When this occurs, a characteristic known as the “leaflesstree pattern” is commonly observed. However, in the earliest stages, the sialography usually shows no change on the sialogram [15].

The treatment of sialomegaly associated with bulimia should be initiated with psychiatric or psychotherapeutic treatment, as treatment for sialomegaly will only be effective if the eating disorder is resolved first. The patient should be aware that the results of the treatment will depend on their purging behaviors, and that relapse of bulimia can lead to recurrence of sialomegaly. There have been cases where sialadenosis spontaneously resolved in the subsequent weeks after cessation of vomiting, but such reports are rare and the sialadenosis usually takes a long time to resolve [3, 6]. Conservative treatment includes the use of sialogogues in the form of sour candy, local heat application and abundant daily hydration [15]. Drug treatment with high doses of pilocarpine hydrochloride (5–15 mg/day) has been described as an effective treatment in some studies, but the results are usually poor [16]. At present, other therapies have been proposed such as glandular chemodenervation with botulinum toxin injections or infusion of triamcinolone, although their effectiveness has not been proved. Surgery is sometimes the only effective

treatment option in case of failure of conservative treatment [15]. Superficial or completely conservative facial nerve parotidectomy is controversial, especially if the eating disorder is not resolved. It is reported that 50 % of people with bulimia nervosa will have recovered completely in 10 years, 33 % will have partially recovered, and 10 and 20 % will still have symptoms [17]. Therefore, it is very important to conduct blood tests and receive the psychiatric assessment confirming that the bulimia has resolved; moreover, after the procedure, the patient should be monitored for at least 2 years, as the recurrence of sialadenosis in patients with active bulimia is very high [18]. Because of the presence of comorbid depression, low rate of complete resolution of the bulimia, the risk associated with surgery, and the absence of definite medical benefits of parotidectomy, some authors recommend that parotidectomy should not be performed in these patients [19]. However, we believe that in select cases such as the case of our patient, surgery can significantly improve comorbid depression due to improvement in the patient's self-image and self-esteem; further, surgery can improve social relationships by eliminating the cosmetic defect involving the parotid swelling and may constitute an incentive for the permanent abandonment of the eating disorder. However, loss of volume in the retromandibular region can also occur after parotidectomy, so surgeons need to inform the patient about this possibility and also assess whether the aesthetic benefits outweigh the aesthetic defects associated with the parotidectomy.

Conclusions

Diagnosing and treating the underlying disorder is important for the management of sialadenosis. In cases of bulimic sialomegaly, conservative measures may not be enough, and parotidectomy may be the only effective treatment option because apart from achieving the main aim of correcting the shape of the face, it can improve adherence to psychiatric treatment by improving the patient's self-esteem.

Compliance with Ethical Standards

Conflicts of interest The researchers declare no conflicts of interest.

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