CASE REPORT

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SIALADENOSIS: A PRESENTING SIGN IN BULIMIA

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Accepted 5 January 1998

Abstract: Background: Sialadenosis refers to noninflammatory, often recurrent, enlargement of the salivary glands, most frequently the parotids, which is almost always associated with an underlying systemic disorder. These include diabetes, alcoholism, malnutrition, anorexia nervosa, and bulimia. It is thought that the various causes 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.

Methods: This paper reports a case of sialadenosis as a presenting sign in bulimia and studies the histologic and electron microscopic features of this disease.

Results: Light microscopy showed acini which appeared to be larger than normal and which were composed of plump pyramidal cells containing prominent zymogen granules. There was less interstitial fat, and the ducts were widely dispersed. Electron microscopy showed the acinar cells to be packed with membrane-limited, dark secretory granules some of which showed moulding of their outlines. Cellular organelles and nuclei were inconspicuous.

Conclusions: Management of sialadenosis depends upon identification of the underlying cause, which must then be corrected. In bulimia, the swellings may be refractory to standard treatment modalities, and parotidectomy may be considered as a last resort to improve the unacceptable aesthetics.


Keywords: sialadenosis; sialosis; bulimia; parotid enlargement

The term “sialadenosis” (sialosis) refers to noninflammatory, often recurrent, enlargement of the salivary glands, most frequently the parotid. These enlargements may be bilateral and are almost always associated with an underlying systemic cause.1 They are usually slow growing and may be associated with pain. The disorder is most often seen between the fourth and seventh decades of life without sex predilection and frequently persists for many years.1,2 There is now general agreement that sialadenosis occurs as a result of a secretory and metabolic dysfunction of the acinar parenchyma.3

The causes of sialadenosis are extremely varied but may conveniently be considered under the headings of endocrine, dystrophic-metabolic, and neurogenic disorders.3–5 Among the most important associated systemic diseases are malnutrition, diabetes mellitus, hypothyroidism, alcoholism, cirrhosis, and anorexia nervosa. In addition, a variety of medications which affect the autonomic nervous system—such as antihypertensive agents, psychotropic drugs, β-adrenergic agents

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and bronchodilators—have also been implicated. Sialadenosis has also been induced in experimental animals using isoprenaline.

The aim of this paper is to report a case of sialadenosis as a presenting sign in bulimia and to study the histologic and electron microscopic features of this disease.

CASE REPORT

The patient was a 21-year-old woman who was initially seen with a 6-week history of bilateral parotid gland swellings which were not painful. The medical history was noncontributory, and she was not on any medications. She gave no history of an eating disorder and, although the patient appeared thin, she indicated that her weight was static. On examination, the patient had diffuse bilateral enlargement of the parotid glands which were nontender to palpation. The submandibular glands were also slightly enlarged. Intraoral examination showed no dental nor mucosal abnormalities. Serologic investigations were normal. Sialography and fine-needle aspiration were performed, both of which were unremarkable.

The patient returned for follow-up 6 months later, and the parotid glands had enlarged significantly. Fine-needle aspiration was again performed and this time showed features compatible with a benign lymphoepithelial lesion. No active treatment was undertaken.

The patient was seen again 2 years later with markedly enlarged parotid glands and complained that these were unsightly and interfered with her work as a photographic model (Figure 1). Bilateral parotidectomies were performed for cosmetic reasons (Figure 2).

Examination of hematoxylin and eosin (H&E) and periodic acid schiff (PAS)–stained sections showed similar features in both glands. The acini appeared to be larger than normal and were composed of plump pyramidal acinar cells which contained abundant, prominent PAS-positive zymogen granules (Figure 3). The nuclei were small, round, and basally situated. As opposed to normal parotid gland, the parenchyma contained fewer, smaller-appearing interspersed adipocytes with only occasional compressed intralobular ducts being noted (Figures 4 and 5). The interstitial fibrous connective tissue contained focal, scattered lymphoid cells.

Tissue from the involved glands as well as a normal parotid control were postfixed with gluteraldehyde and osmium tetroxide and prepared for transmission electron microscopy. In the affected glands, the acinar cells contained numerous, densely packed, dark membrane-limited secretory granules (Figure 6) which were of a size similar to the granules in the control tissue (Figure 7). The granules were so closely packed that...
molding of their outlines had occurred. The nuclei and other cellular organelles were not easily identifiable (Figure 6). In contrast, the granules in the control tissue were far fewer in number and were round in shape, and the nuclei and other cellular organelles could readily be seen (Figure 7). A diagnosis of sialadenosis was made.

Further questioning of the patient at this time elicited a long-standing history of bulimia.

DISCUSSION

In the differential diagnosis of bilateral recurrent parotid swellings, conditions such as sialadenosis, sialadenitis, sarcoidosis, and Sjögren’s syndrome need to be considered. Evaluation of the clinical and medical history in association with sialography, serology, fine-needle aspiration, and incisional labial gland or parotid biopsy may be necessary to reach a definitive diagnosis.2,8

As already pointed out, the causes of sialadenosis are extremely varied, but it is thought that they result in a common pathogenetic effect in that they produce a peripheral autonomic neuropathy which is responsible for disordered salivary metabolism and secretion.1,2,5 Donath and Seifert9 have provided ultra-structural evidence in support of this theory. Their studies showed degenerative changes both in myoepithelial cells and in autonomic nervous system postganglionic sympathetic neurones. In the former, these consisted of nuclear pyknosis, fat vacuoles, and homogeneous condensation zones of the cytoplasm, whereas in the latter, there was a loss of neurosecretory granules, destruction of the mitochondrial...
dria, hydropic swelling of the axoplasm, and a terminal axolysis. It appears as if these cellular changes may result in both a lengthening of the storage phase of the secretory granules and an arrest in protein synthesis by the acinar cells, both processes producing accumulation of granules and marked enlargement of acini.\(^2\) The diameter of the acini may increase to two or three times that of normal, reaching a size ranging from 50 to 100 \(\mu\)m.\(^2\)

Three histologic patterns of sialadenosis are recognized: granular, honeycomb, and mixed. These differences are determined solely by the optic density of the secretory granules.\(^2\) Electron microscopy has shown that the granules may be either dark (protein-rich) or clear (protein-weak).\(^9\) In our case, only dark granules were observed.

Sialography can play an important role in the diagnosis of sialadenosis, particularly in the later stages of the disease, when changes are caused by acinar swelling with compression of the proximal ducts. This results in a characteristic “leafless-tree pattern.”\(^4,10,11\) In the early stages of the disease, there may be no sialographic changes, as occurred in our case.

Anorexia nervosa and bulimia are complex psychological eating disorders which result in variable and often serious manifestations including metabolic disturbances, hirsutism, electrolyte imbalances, hypotension,\(^12\) the loss of tooth structure,\(^13–16\) increased frequency of dental caries, gingivitis,\(^16\) and sialadenosis.\(^17–19\) The exact etiology and pathogenesis of sialadenosis in these eating disorders are unknown but are likely to be multifactorial in nature. The incidence of parotid enlargement in bulimia varies from 10% to 66%.\(^20\)

The management of sialadenosis depends upon the possible identification of a cause, which must then be corrected. In many instances, this will result in complete resolution. However, many cases, especially those with an endocrine or neurogenic basis, are refractory to treatment and show no improvement, even when the basic underlying systemic disorder is adequately controlled.\(^1,2\) Treatment using \(\beta\)-receptor blockers has not resulted in appreciable improvements in animal experiments.\(^2\)

In bulimia, cessation of vomiting is generally associated with resolution of the parotid swelling, however, this may not be immediate. The standard treatment modalities include heat application to the swollen glands, salivary substitutes, and the use of sialogogues. There have however been cases reported in the literature that have failed to respond to these therapeutic interventions.\(^18,21\) In two such cases, pilocarpine hydrochloride drops were administered orally at a dosage of 1.25 to 5.0 mg/day. This resulted in a marked diminution in the size of the parotid glands enlargement.\(^18\)

In those cases of sialadenosis which prove refractory to treatment, parotidectomy may be considered as a last resort to improve unacceptable aesthetics.\(^2,18\)

**Acknowledgments.** The authors are grateful to Mrs H. Ali and L. v.d. Walt for their technical assistance.

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