

Chronic Sclerosing Sialadenitis (Küttner Tumor) in the sublingual gland: unusual manifestation related to partial edentulism and chronic masticatory trauma.

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Received: 05/08/2010
Accepted: 06/09/2010

Pinheiro TN. Chronic Sclerosing Sialadenitis (Küttner Tumor) in the sublingual gland: unusual manifestation related to partial edentulism and chronic masticatory trauma. J Clin Exp Dent. 2011;3(2):e177-9. <http://www.medicinaoral.com/odo/volumenes/v3i2/jcedv3i2p177.pdf>

Article Number: 50369 <http://www.medicinaoral.com/odo/indice.htm>
© Medicina Oral S. L. C.I.F. B 96689336 - eISSN: 1989-5488
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Abstract

Chronic sclerosing sialadenitis or Küttner tumor is an unusual chronic inflammatory disease of the salivary gland that mimics a malignant neoplasm clinically because of presentation as a hard mass. The diagnosis can only be made histologically and is an underrecognized entity. Recent studies have shown important features that characterizes the disease mainly as an autoimmune reaction. The aim of this work is to report a case of a 40-year-old man, presenting with a three-year history of a painless, moderate sublingual mass related to a partial edentulism of the teeth 36 and 37. Functional evaluation revealed an awkward misplacement of the mass into the edentulous site. Clinical and radiographic procedures revealed a decreased salivary flow and no signs of remarkable pain or sialolithiasis. Sublingualectomy was performed and histopathological examination confirmed the presence of non-obstructive chronic sclerosing sialadenitis of the sublingual gland. The possible autoimmune reaction triggered by hidden (sequestered) antigens exposed by chronic masticatory trauma is discussed.

Key words: Chronic sclerosing sialadenitis, Küttner tumor.

Introduction

Chronic sclerosing sialadenitis is a relatively uncommon and underrecognized cause of salivary gland enlargement that characteristically affects the submandibular salivary gland. First described by Küttner in 1896 (1), because of its clinical similarity to a salivary gland neoplasm, this disease has been referred to as Küttner tumor and is classified as a tumorlike lesion (2, 3). The aim of this work is to report a case of chronic sclerosing sialadenitis of the sublingual gland related to partial edentulism and chronic masticatory trauma.

Histologically the different types of chronic sialadenitis are characterized by acinar atrophy, lymphocytic infiltrates and progressive fibrosis. Clinically relevant factors such as localization of the major or minor salivary glands, aetiological factors (bacterial, viral, radiation-related, immunological factors), the course of the disease (acute, chronic, recurring) and the patient's age and sex determine the classification of the sialadenitis (4-6).

According to Seifert and Donath (4) the lesion may evolve through four histological stages.

1. Focal chronic inflammation is present with periductal lymphocytic infiltration and dilated ducts containing inspissated secretion.
2. Marked diffuse lymphocytic infiltration and more severe periductal fibrosis are apparent. Duct system exhibits hyperplasia of the ductal epithelium with occasional epimyoeplithelial islands. Periductal lymphoid follicles are well-developed. There is fibrosis in the centers of the lobules and atrophy of acini.
3. Reduction of secretory gland parenchyma, secondary lymphoid follicle formation with reactive germinal centers and extensive fibrosis, and ductal proliferation are present. Squamous and goblet cell metaplasia are apparent in the ductal system.
4. Destruction of the lobular architecture is apparent, and sclerosis-cirrhosis of the gland occurs.

Case report

A 40-year-old man presented with a three-year history of a painless, moderate sublingual mass related to a partial edentulism of the teeth 36 and 37. The sublingual mass occupied the site of the missing teeth during speech and masticatory function. Milking and palpation procedures, revealed a decreased salivary flow and no signs of remarkable pain or sialolithiasis. Oclusal radiography procedure did not revealed sialolithiasis whatsoever. Sublingualectomy was performed, without any particular complications though the following 3 month of follow-up period Fig. 1 and 2.

Histologically, the normal lobular architecture was preserved. The firm area noted on gross examination exhibited patchy infiltrates of lymphocytes, plasma cells, and lymphoid follicles. This inflammatory infiltrate was associated with acinar atrophy in many areas. The ducts



Fig. 1. Clinical aspects of the sublingual mass. Note that the normal anatomy of the left side of the mouth floor is altered. Functional evaluation revealed an awkward misplacement of the mass into the edentulous site.



Fig. 2. Palpation and milking procedures revealed decreased salivary flow, as well as absence of sialolithiasis.

appeared dilated; some containing inspissated secretions, and was surrounded by fibrous tissue. Fibrosis was noted predominantly in the periductal region with foci extending into the surrounding interlobular septa. Foci of squamous cell metaplasia were noted in the dilated ducts Fig. 3.

Discussion

Chronic sclerosing sialadenitis is characteristically of the submandibular gland, but multiglandular involvement has been described (7). Seifert and Donath (4) hypothesized that initially a functional abnormality leads to inspissated secretion in the small ducts, leading to destruction of the epithelial structure of the involved gland. Tiemann et al. (5) determined the phenotype of the immunocompetent cells in chronic sclerosing sialadenitis of the submandibular gland. This disease is characterized by an abundance of CD8-positive T cells and cytotoxic destruction of glandular epithelial cells with

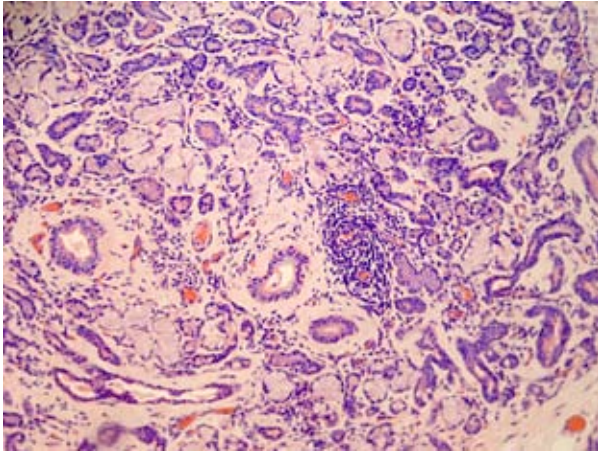


Fig. 3. Microscopic features of chronic sclerosing sialadenitis - Küttner tumor.

features of an autoimmune process. A recent study with western population found that chronic sclerosing sialadenitis belongs to the spectrum of IgG4-related diseases (8).

Since Küttner(1)described the condition, more than a century ago, chronic sclerosing sialadenitis has remained as an underrecognized condition. Recently, with a better understanding of the cell lineage of the inflammatory infiltrate present in the lesion, the autoimmune explanation of the process has gained particular highlight (5-8). A reasonable consideration could be stated based on an autoimmune reaction, triggered by hidden or sequestered antigens. In the presented case, the long term masticatory trauma caused by the displacement of the sublingual gland into the site of the missing teeth 36 and 37 probably was able to begin an inflammatory reaction and release hidden antigens. Another probable cause may be attributed to common or so-called cross-reactive antigens between micro-organisms and mammalian tissues. The coexistence of chronic sialadenitis in a patient with pleomorphic adenoma of the parotid, reported elsewhere (9) points to the same direction. Therefore, many aetiological factors related to common chronic sialadenitis could potentially trigger a chronic sclerosing sialadenitis event.

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