

<https://doi.org/10.17221/109/2018-VETMED>

Sclerosing sialadenitis of the sublingual salivary glands in a cat

V. IPEK^{1*}, G. CECEN AYALP², M.B. AKGUL³, M. AKBALA², G. SONMEZ²

¹Faculty of Veterinary Medicine, Mehmet Akif Ersoy University, Burdur, Turkey

²Faculty of Veterinary Medicine, Uludag University, Bursa, Turkey

³Faculty of Veterinary Medicine, Siirt University, Siirt, Turkey

*Corresponding author: volkanipek@mehmetakif.edu.tr

ABSTRACT: In this report, sclerosing sialadenitis of the sublingual salivary glands is described in a three-year-old, male, mixed breed cat. Sublingual masses were dissected and removed under dissociative anaesthesia. The patient had recovered completely at two months post-surgery. The two soft and pale red masses were 1.5 × 1 × 0.5 cm in size. Histologically, it was observed that the masses were covered with stratified squamous epithelium containing large numbers of salivary glands and ducts in the collagen-rich loose connective tissue. Scattered lymphocyte infiltrations were observed in the connective tissue. Mild to severe lymphocyte infiltration was seen between salivary glands. Some glands exhibited severe fibrosis, epithelial destruction and atrophy. To the best of our knowledge, this is the first report of sclerosing sialadenitis in a cat. Chronic sclerosing sialadenitis (Kuttner's tumour) is a condition that has been defined in humans. Histological findings similar to those of Kuttner's tumour were observed in this case, and dense plasma cell infiltrations suggested immune-mediated plasmacytic disease, which has been reported as a potential aetiology of Kuttner's tumour. Corticosteroid therapy was not attempted, and the patient was treated successfully using surgical excision.

Keywords: feline; Kuttner's tumour; salivary gland

Chronic sclerosis sialadenitis was first described by Kuttner in 1896 in humans and the lesion was reported to be associated with sialolithiasis (Kuttner 1896). Although it is often referred to as Kuttner's tumour (KT) because of its clinical appearance, it is actually an inflammatory disease of the salivary gland (Pinheiro 2011; Pandarakalam et al. 2013). Differential histological features of KT include periductal sclerosis, lymphocytic infiltration, lymphocytic follicles, atrophic ascites and fibrosis (Pandarakalam et al. 2013). In the classification of salivary gland tumours of the World Health Organization (WHO), KT is in the category of tumour-like lesions (Seifert 1992). Sialoliths are frequently observed in KT and are considered to be the most common aetiological factor. Other predicted aetiologies include secretory disorders, infectious agents and immunologic responses (Seifert and Donath 1977; Harrison et al. 1997; Chan 1998;

Tiemann et al. 2002). It has also been suggested that KT may be associated with immune-mediated processes (Abe et al. 2009). Salivary gland tumours, necrotising sialadenitis, salivary mucocele and sialadenosis have been reported in salivary gland diseases of cats (Boydell et al. 2000; Hammer et al. 2001; Brown et al. 2004; Langley-Hobbs 2014). According to the authors' knowledge, there is no report of sclerosing sialadenitis in cats.

In this case report, we report bilateral tumour-like formations in the sublingual salivary glands of a cat, which are similar to chronic sclerosing sialadenitis in humans.

Case description

A three-year-old, male, mixed breed cat presented with breathing discomfort and dysphagia. In the

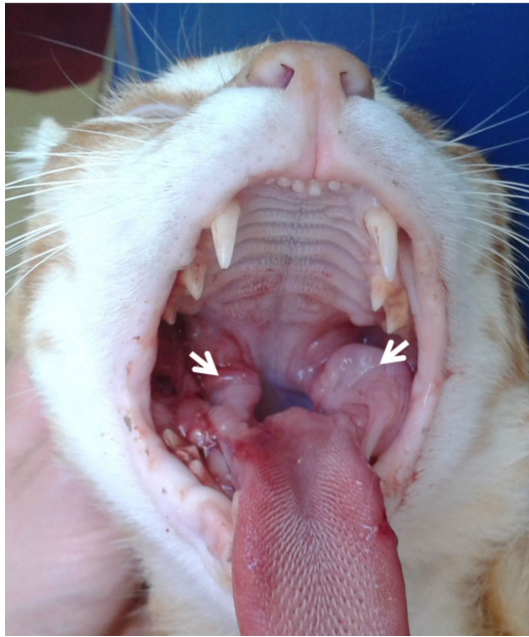


Figure 1. Enlarged sublingual salivary glands (arrows) on the first examination

clinical examination, bilateral soft and non-painful masses extending from the base of the tongue to the molar teeth were detected (Figure 1). These masses covered the tongue and pharynx during swallowing. Bloody saliva, dental calculus on the third premolar teeth of the maxilla and focal mucosal erythematous areas at the level of both mandibular and maxillary molar teeth were observed. Considering the anatomical location of the masses, sublingual gland hyperplasia was diagnosed clinically and operative treatment was performed. In preoperative period, the cat received cephalosporin (10 mg/kg *i.m.* q8h for six days), tolfenamic acid (4 mg/kg *s.c.* q24h for two days) and 10% iodine glycerine solution (twice a day topically). After this treatment, no reduction in the size of the masses was observed. Surgical removal of the masses was scheduled for six days later, and the sublingual masses were dissected and removed under dissociative anaesthesia. The patient had recovered completely at two months post-surgery (Figure 2).

The masses were 1.5 × 1 × 0.5 cm in size and were soft with a pale red colour. Touch imprints were made from the cross sections of masses and slides were stained with Hemacolor® (Merck Millipore). Cytological examination showed multiple epithelial cells with neutrophil leukocytes and a small number of lymphocytes (Figure 3). No bacteria were found free or within the neutrophil cytoplasm.



Figure 2. Full recovery after surgery

For histopathological examination, the two masses were fixed in 10% formaldehyde and embedded in paraffin following routine follow-up procedures; 5-µ-thick sections were taken. Slides were stained with haematoxylin-eosin solution and examined under a light microscope. Histopathological examination revealed that the masses were covered with stratified squamous epithelium and contained salivary glands and draining ducts within collagen-rich loose connective tissue. A large number of plasma cells and, less commonly, lymphocyte infiltrates were found scattered throughout the connective

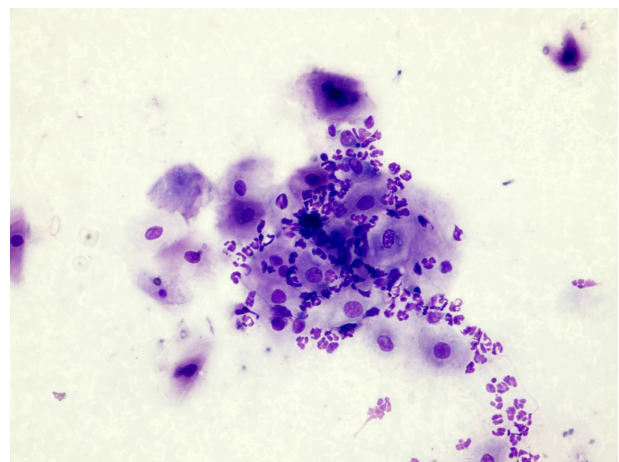


Figure 3. Epithelial cells and neutrophil leukocytes stained with Hemacolor, × 200

<https://doi.org/10.17221/109/2018-VETMED>

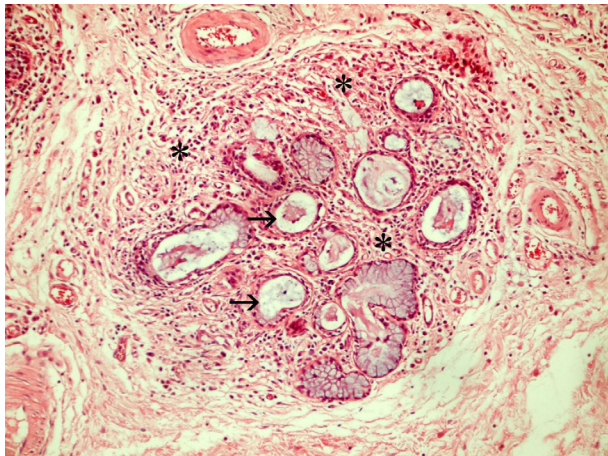


Figure 4. Fibrosis and mononuclear cell infiltrations between glands (asterisks) and destruction of some gland epithelium (arrows), Haematoxylin-Eosin, $\times 200$

tissues. Some glandular areas showed lymphocytic-plasmacytic infiltrations, fibrosis, epithelial destruction and atrophy (Figures 4 and 5).

DISCUSSION AND CONCLUSIONS

KT in salivary glands is underreported, and is usually found in the submandibular salivary glands as well as in the parotid glands and sublingual salivary glands (Tiemann et al. 2002; Pandarakalam et al. 2013). In our case, the lesions were found bilaterally in the sublingual salivary glands. Histologically, there are different types of KT characterised by acinar atrophy, lymphocytic infiltrates and progressive

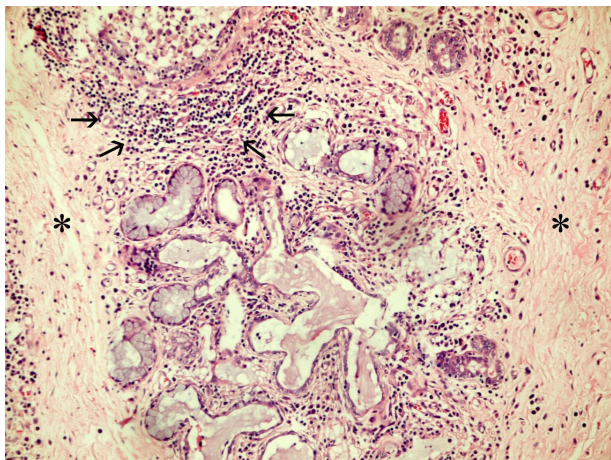


Figure 5. Collagen-rich fibrous tissue proliferation (asterisks) and lymphocytic-plasmacytic infiltrations (arrows) between glands, Haematoxylin-Eosin, $\times 200$

fibrosis (Tiemann et al. 2002). Seifert and Donath classified the lesions into four different histological categories (Seifert and Donath 1977). In the first category, focal sialadenitis is characterised by periductal lymphocytic infiltration and focal chronic inflammation with dilate ducts containing thickened secretions. The second category includes diffuse lymphocytic sialadenitis characterised by fibrosis in the centres of the lobules and atrophy in the acinus with marked diffuse lymphocytic infiltration, more severe periductal fibrosis, hyperplasia of the duct epithelium and well-developed periductal lymphoid follicles. In the third category, chronic sclerosing sialadenitis is characterised by a decrease in secretory gland parenchyma, secondary lymphoid follicle formation with reactive germinal centres, severe fibrosis, ductal proliferation and salivary gland sclerosis with marked squamous and goblet cell metaplasia in the ductal system. In the last category, chronic progressive sialadenitis is characterised by sclerosis and cirrhosis of glands and the marked destruction of lobular structures in the cortex. In our case, sclerosing sialadenitis was diagnosed in accordance with the description of lesions classified into the third category and was characterised by atrophy and destruction of some glands, secondary lymphoid follicle formations with active germinal centres and decreased salivary gland parenchyma due to severe fibrosis between the glands.

With respect to aetiology, it has been stated that KT is most related to sialolithiasis, and less commonly to other aetiologies include secretory disorders, infectious agents and immunologic responses (Seifert and Donath 1977; Harrison et al. 1997; Chan 1998; Tiemann et al. 2002; Kitagawa et al. 2005; Abe et al. 2009). There are some reports of necrotising sialadenitis in cats and dogs that are thought to be a consequence of ischaemic lesions due to vascular damage, although the precise aetiology is still unclear (Mawby et al. 1991; Brooks et al. 1995; Brown et al. 2004). Salivary gland enlargements called sialadenozis together with a lack of abnormal findings in the histological examination have also been reported (Boydell et al. 2000). We did not observe any sialoliths macroscopically or microscopically, suggesting that the lesions in this case may have had other aetiologies. Cytological and histopathological examination did not reveal the presence of bacteria. No cystic formations showing any obstruction were found. However, the finding of plasma cell infiltrations suggests that the lesions may have

<https://doi.org/10.17221/109/2018-VETMED>

developed due to immune-mediated plasmacytic disease. Indeed, Kitagawa et al. (2005) and Abe et al. (2009) found the presence of IgG4-positive plasma cell infiltrates in two cases of sclerosing sialadenitis and reported complete recovery with corticosteroid treatment. In this presented case, the masses were removed surgically and full recovery was achieved but corticosteroid therapy was not attempted.

In contrast to all previously described manifestations of disease and tumours in this condition, histologically we observed lymphoid foci containing intense plasma cell infiltrations and severe fibrosis in the salivary glands. This appearance differs from that of the previously reported salivary gland lesions in cats and dogs. On the other hand, the macroscopic and microscopic similarities with the appearance of KT in humans support the idea that the lesions we described here may be similar to those in humans and have similar aetiology. Immune-mediated plasmacytic disease has been suggested as an aetiology of KT by some researchers (Kitagawa et al. 2005; Abe et al. 2009) due to the dense plasma cell infiltrations that are observed. Sclerosing pancreatitis and other immune-mediated plasmacytic diseases have been reported in some cases. However, in some other reports it is also stated that no other disorders are observed (Kitagawa et al. 2005; Abe et al. 2009). In our case, there were no other complaints which would have been suggestive of any disturbance. In conclusion, this case of sclerosing sialadenitis is similar to KT in terms of histopathological findings and according to the authors' knowledge, this is the first report of this condition in a cat.

REFERENCES

- Abe T, Sato T, Tomaru Y, Sakata Y, Kokabu S, Hori N, Kobayashi A, Yoda T (2009): Immunoglobulin G4-related sclerosing sialadenitis: report of two cases and review of the literature. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 108, 544–550.
- Boydell P, Pike R, Crossley D (2000): Presumptive sialadenitis in a cat. *Journal of Small Animal Practice* 41, 573–574.
- Brooks DG, Hottinger HA, Dunstan RW (1995): Canine necrotizing sialometaplasia: a case report and review of the literature. *Journal of the American Animal Hospital Association* 31, 21–25.
- Brown PJ, Bradshaw JM, Sozmen M, Campbell RH (2004): Feline necrotising sialometaplasia: A report of two cases. *Journal of Feline Medicine and Surgery* 6, 279–281.
- Chan JK (1998): Kuttner tumor (chronic sclerosing sialadenitis) of the submandibular gland: an underrecognized entity. *Advances in Anatomic Pathology* 5, 239–251.
- Hammer A, Getzy D, Ogilvie G, Upton M, Klausner J, Kisseberth WC (2001): Salivary gland neoplasia in the dog and cat: survival times and prognostic factors. *Journal of the American Animal Hospital Association* 37, 478–482.
- Harrison JD, Epivatianos A, Bhatia SN (1997): Role of microoliths in the aetiology of chronic submandibular sialadenitis: A clinicopathological investigation of 154 cases. *Histopathology* 31, 237–251.
- Kitagawa S, Zen Y, Harada K, Sasaki M, Sato Y, Minato H, Watanabe K, Kurumaya H, Katayanagi K, Masuda S, Niwa H, Tsuneyama K, Saito K, Haratake J, Takagawa K, Nakanuma Y (2005): Abundant IgG4-positive plasma cell infiltration characterizes chronic sclerosing sialadenitis (Kuttner's tumor). *The American Journal of Surgical Pathology* 29, 783–791.
- Kuttner H (1896): About inflammatory tumours of the submaxillar salivary gland (in German). *Brunns' Beitrage Zur Klinischen Chirurgie* 15, 815–828.
- Langley-Hobbs S (2014): The head and neck. In: Langley-Hobbs SJ, Demetriou JL, Ladlow JF (eds): *Feline Soft Tissue and General Surgery*. Saunders. 571–586.
- Mawby DI, Bauer MS, Lloyd-Bauer PM, Clark EG (1991): Vasculitis and necrosis of the mandibular salivary glands and chronic vomiting in a dog. *The Canadian Veterinary Journal* 32, 562–564.
- Pandarakalam C, Goebel WM, Seyer B (2013): Chronic sclerosing sialadenitis or Kuttner's tumor associated with a giant sialolith: A case report. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology* 115, e38–e40.
- Pinheiro TN (2011): Chronic sclerosing sialadenitis (Kuttner tumor) in the sublingual gland: Unusual manifestation related to partial edentulism and chronic masticatory trauma. *Journal of Clinical and Experimental Dentistry* 3, 177–179.
- Seifert G (1992): Tumour-like lesions of the salivary glands. The new WHO classification. *Pathology, Research and Practice* 188, 836–846.
- Seifert G, Donath K (1977): On the pathogenesis of the Kuttner tumor of the submandibular gland – analysis of 349 cases with chronic sialadenitis of the submandibular. *HNO* 25, 81–92.
- Tiemann M, Teymoortash A, Schrader C, Werner JA, Parwaresch R, Seifert G, Kloppel G (2002): Chronic sclerosing sialadenitis of the submandibular gland is mainly due to a T lymphocyte immune reaction. *Modern Pathology* 15, 845–852.

Received: July 29, 2018

Accepted after corrections: September 27, 2018