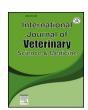
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Case Report

Lipomatosis of the parotid salivary gland: 2 case reports and review of the literature on fat-containing salivary gland lesions



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ABSTRACT

Two cases of lipomatosis of the parotid salivary gland in two dogs are reported. Both dogs were presented with a slow growing unilateral non-painful mass in the parotid region. The mass measured 12 and 15 cm on the longest axis for case 1 and 2 respectively. Initial incisional biopsies were suggestive of infiltrative lipoma or salivary gland adenocarcinoma. Case 2 underwent head radiographs that showed a soft tissue opacity on the area of the right parotid gland followed by a computed tomography that showed a multi-lobulated infiltrative mass occupying the right parotid gland. Both cases underwent total parotidectomy with an excisional biopsy intent. Surgery on case 1 included total ear canal ablation (TECA). Histology revealed lipomatosis of the parotid gland with a similar admixture of adipocytes and well-differentiated salivary acini on both cases. No recurrence was reported by 4 years for case 1 and by 3 years for case 2. Case 1 developed long-term right-sided facial paralysis and palpebral reflex deficit due to damage of the facial and trigeminal nerve during surgery. The cases are described and the veterinary literature is reviewed in view of the most updated information available in human medicine on fat-containing salivary gland lesions. Considering this literature review, authors recommended these two cases to be classified as lipomatosis of the salivary gland. In conclusion, fat-containing salivary gland lesions should be considered in the list of differential diagnoses in dogs with an enlarged salivary gland and seem to be associated with an excellent prognosis after surgical excision.

1. Introduction

Primary salivary neoplasia is rare in dogs and clinical signs are usually non-specific. In most cases the neoplasm is malignant with adenocarcinoma being the most common histological subtype. Other primary salivary malignancies in dogs include osteosarcoma, mast cell tumour and malignant fibrous histiocytoma. Enlargement of the salivary gland in dogs can also be caused by benign neoplastic and nonneoplastic processes such as salivary adenoma, sialoadenosis, salivary cyst and salivary infarct [1–8].

In human patients the salivary gland can also be enlarged due to fatcontaining lesions. These are uncommon and include sialolipoma, salivary lipomatosis, lipoadenoma, angiolipoma and adenoma with lipometaplasia [9–12]. In dogs there are few reported cases of fat-containing salivary gland lesions and most of these have been classified as either sialolipoma or salivary lipomatosis [7,13]. Sialolipoma and salivary lipomatosis are histologically similar and in human medicine the distinction between these two entities is based on the presence (sialolipoma) or absence (salivary lipomatosis) of a capsule at the periphery of the lesion [11].

Unfortunately, this diagnostic criterion has not been strictly applied in all veterinary publications, leading to some confusion in the classification of these lesions in dogs.

The authors here describe two cases of parotid lipomatosis and discuss the veterinary literature in view of the most updated information available in human medicine for fat-containing salivary gland lesions.

2. Cases presentation

Case 1: A 6-year-old neutered male Border Collie dog was referred to the "Institution 1" with an over 8-month history of a progressively enlarging soft tissue mass in the right parotid region. The mass had been biopsied twice (incisional biopsies) 7 months and 4 weeks before

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Fig. 1. A: Photograph of case 1 showing a well-defined soft tissue mass on the right parotid region. B: Photograph of case 2 showing a right retromandibular soft tissue mass.

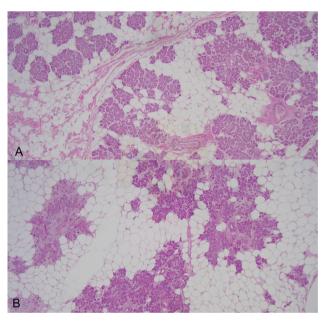


Fig. 2. A: Microphotograph of excised tissue from case 1 showing an admixture of mature adipocytes interspersed with well-differentiated salivary acini and few salivary ducts. Haematoxylin and eosin stain, $4 \times$. B: Microphotograph of excised tissue from case 2 showing a similar admixture of adipocytes and well-differentiated salivary acini. Haematoxylin and eosin stain, $4 \times$.

referral. Histology of the first biopsy was suggestive of an infiltrative lipoma whilst the second examination suggested a well-differentiated salivary gland adenocarcinoma. Both biopsies were performed in a private veterinary diagnostic laboratory. Slides were not available for review.

Upon presentation, a well-defined approximately 12 cm in diameter dense soft tissue mass was observed in the right parotid region (Fig. 1A). The mass was mobile and non-painful. On palpation, the mandibular lymph nodes were normal. Haematology and biochemistry were unremarkable. Inflated lateral and ventro-dorsal thoracic radiographs and abdominal ultrasound were unremarkable.

The owners declined further investigations and elected to proceed for exploratory surgery.

Exploratory surgery and total parotidectomy was performed using a right lateral approach to the parotid region. The mass was adherent to the right external ear canal and therefore, Total Ear Canal Ablation (TECA) of the right ear was performed. Part of the facial and trigeminal nerves had to be sacrificed. Right-sided facial paralysis and deficit of the palpebral reflex developed post-operatively.

Histological examination of the excised tissue performed by a board-certified pathologist showed expansion of the salivary gland by a non-capsulated mass composed of mature adipose tissue interspersed with well-differentiated salivary acini and ducts (Fig. 2A).

Adipocytes occupied approximately 50% of the mass. Given the lack of a capsule, this fat-containing lesion was classified as salivary lipomatosis.

Four years after surgery there was no evidence of recurrence or systemic disease. The facial paralysis and deficit on palpebral reflex persisted, although the owners reported the dog having a normal life.

Case 2: A 10-year old entire male English Setter was referred to "Institution 2" with a 3 years history of a progressively enlarging right retromandibular soft tissue mass. Two and half years before referral, incisional biopsy revealed glandular atrophy and fibrosis with mild chronic inflammation. Slides were not available for review.

Upon presentation, an approximately 15 cm in diameter dense soft tissue mass was observed in the right retromandibular region (Fig. 1B). Physical examination was otherwise unremarkable.

Dorso-ventral radiographs of the head showed a well-defined soft tissue opacity in the parotid area with no evidence of osteolysis (Fig. 3). Computed Tomography (CT) scan of the affected region showed a severe but not infiltrative irregular lobulation of the right parotid gland (Fig. 3).

Total parotidectomy was performed using a lateral approach with an incision parallel and caudal to the mandibular branch, over the vertical canal. The mass was dissected, sparing the horizontal canal and the facial nerve. A smaller adjacent mass was found during surgery and also excised from the right retro-parotid area. The right mandibulary salivary gland was also excised.

Histological examination of the excised tissue performed by a board-certified pathologist showed a non-capsulated expansion of the gland composed of lobules of mature adipocytes intermingled with lobules of salivary acini and ducts (Fig. 2B). Adipocytes occupied approximately 60% of the mass. There was no evidence of concurrent inflammation or fibrosis. Given the lack of a capsule, this fat-containing lesion was classified as salivary lipomatosis. Histology of the right mandibulary gland was unremarkable.

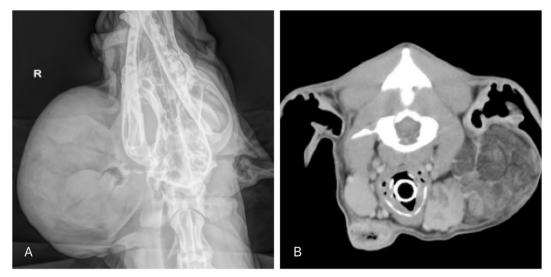


Fig. 3. A: Radiographic image of the head of the case 2 showing a soft tissue opacity on the right parotid region and no evidence of osteolyis. B: Transverse plan image of computerized tomography from the head of the case 2 showing a multi-lobulated infiltrative mass occupying the right parotid gland. No evidence of osteolysis was present. The right mandibulary gland had an heterogenous appearance but histology showed normal salivary gland tissue. On the left side, just ventral to the left mandibulary salivary gland there was a soft tissue opacity structure with air-filled, of unknown significance, that had cytological features of inflammation and disappeared with an antibiotic course.

The dog died 3 years later from non-related causes and no local recurrence was evident.

3. Discussion and review of literature

Fat-containing salivary gland lesions are uncommon in human patients and only few cases have been reported in dogs (Table 1). In dogs most of these lesions affect the parotid or submandibular salivary gland. There is a single report of a salivary angiolipoma (benign tumour composed of adipose tissue and blood vessels) within the parotid gland [4], but all the other canine fat-containing salivary gland lesions (14 cases) have been described as consistent with either sialolipoma (3 cases) or salivary lipomatosis (also known as lipomatous/fatty infiltration of the salivary gland) (11 cases). Sialolipoma and salivary lipomatosis are histologically similar as they both consist of mature adipose tissue intermingled with well-differentiated salivary gland tissue. In human medicine the distinction between these two histopathological entities has been recently detailed [9]. The most distinctive feature is the peripheral capsule, which is present in sialolipoma and absent in lipomatosis. The 3 cases described as sialolipoma in dogs were reported to have the typical peripheral capsule [7,8]. However, a peripheral capsule was also present in a case that was classified as lipomatosis [14] and that, in the authors' opinion, would need to be reclassified as sialolipoma. In addition, there are 3 published canine cases of lipomatosis/fatty infiltration of the salivary gland for which it is not described whether a capsule was present or not [6].

We believe that a strict application in veterinary medicine of the human diagnostic histological criteria for classifying fat-containing salivary gland lesions would allow an easier comparison with the human literature and to more precisely determine the incidence of these lesions in dogs.

The reason why sialolipoma and salivary lipomatosis in human medicine are considered as separate entities is because sialolipoma is, by definition, a neoplastic process, whereas lipomatosis is suspected to represent a non-neoplastic condition, possibly secondary to an underlying obstructive or inflammatory disease of the gland [9]. Interestingly, in one of the two canine cases of salivary lipomatosis presented here mild inflammation and glandular fibrosis/atrophy were noticed on an incisional biopsy performed two and a half years before the surgery and these changes might have favoured the development of the lipomatosis

In human medicine the treatment of choice is surgical excision for sialolipoma as well as salivary lipomatosis and after surgery recurrence has not been described [12]. This is in agreement with the clinical course observed for the two canine patients described here and for the other canine cases documented in the literature.

Although sialolipoma and salivary lipomatosis appear to be associated with an excellent prognosis after surgery, the diagnosis is challenging, as incisional biopsies are frequently misleading. Given the normal appearance of the adipose tissue and salivary gland tissue, cytology is also unlikely to be diagnostic. An increased awareness in veterinary medicine of the existence of these lesions could help pathologists and clinicians in recognizing them, especially when repeated incisional biopsies and fine needle aspirates keep showing a combination of normal adipocytes and salivary gland cells. Advanced imaging such as Computed Tomography or Magnetic Resonance Imaging scan could also be considered for sample guidance.

In conclusion, fat-containing salivary gland lesions should be considered in the list of differential diagnoses in dogs with an enlarged salivary gland and are associated with an excellent prognosis after surgical excision. Amongst these lesions, lipomatosis appears to be the most common, followed by sialolipoma. A strict application in veterinary medicine of the human diagnostic histological criteria for classifying fat-containing salivary gland lesions is advisable.

Competing of interests

No conflict of interests have been declared.

(continued on next page)

Table 1

Diagnosis in authors opinion	Salivary lipomatosis	Salivary lipomatosis	Salivary lipomatosis	Salivary lipomatosis	Salivary lipomatosis	Salivary lipomatosis	Salivary Iipomatosis	Salivary lipomatosis	Uncertain	Uncertain	Uncertain	Angiolipoma	Sialolipoma	Sialolipoma
												An	Sia	Sia
Original diagnosis	Salivary gland lipomatosis	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary glands	Lipomatous infiltration of salivary olands	Fatty infiltration/degeneration	Fatty infiltration/ degeneration	Fatty infiltration/ degeneration	Infiltrative angiolipoma	Sialolipoma	Sialolipoma
Histological findings	Salivary gland with a marked fatty infiltration separating the individual glands into groups of various sizes sparing the piece that appeared grossly normal.	Extensive replacement of parenchymal tissue by irregular lobules of uniform, well-differentiated fat cells. Portions of	persisting glandular tissue had the typical histological appearance of salivary gland. None of the tissues appeared to have a	distinct capsule defining the margins of the gland or the fatty tissue within the gland.	ò				NS	NS	NS	Multifocal to coalescing single or clusters of blood-filled vessels lined by flattened endothelial cells with narrow, elongated, basophilic nuclei. These regions were embedded in adipose tissue with multifocal areas of intervascular remnants of differentiated serous salivary oland tissue with multifocal small dures	Lobules of mature adjoose tissue with smaller amounts of salivary tissue and ducts, surrounded by a fibrous connective tissue capsule	Presence of mature adipocytes admixed with foci of salivary tissue composed of both ductular and actinar elements. The mass was partially surrounded by a fibrous capsule and there was normal salivary gland and a linear area of fibroplasia admixed with neovascularisation
Follow up	NS	NS	NS	NS	NS	NS	NS	NS	NS	NS	NS	Continued growing- in 14 months has grown 30%	No regrowth after 6 months	No recurrence after 7 months
Treatment	Gland excision	Gland excision	Gland excision	Gland excision	Gland excision	Gland excision	Gland excision	Gland excision	NS	NS	NS	No treatment	Mass excision	Gland excision
Size	7 × 6 × 5 cm	$2.5\times1.5\times1~\text{cm}$	$7 \times 5 \times 3$ cm	$5 \times 4 \times 2$ cm	Twice normal	2 cm	$2 \times 1 \text{cm}$	1.5 cm	NS	NS	NS	10 × 5 × 5 cm	5×6 cm (mass) plus 1.5×0.5 cm pedicle	SN
Duration of swelling/ clinical signs	1 year	Slow growing mass	Slow growing over 4 months	Firm swelling increasing in size	Slow growing mass	Mass more than 4 weeks duration	Dysphagia	Salivary gland swelling	NS	NS	NS	Growing over 18 months		Three weeks
Salivary gland affected	Mandibulary (side unknown)	Left parotid	Parotid (side unknown)	Right Parotid	Right submandibular	Left submandibular	Right submandibular	Submandibular (side unknown)	NS	NS	NS	Parotid (side unknown)	Minor salivary gland close to right tonsillary fossa (side unknown)	Right parotid
Breed	Beagle	Labrador	Retriever	Schnauzer	ESS	Crossbreed	Terrier crossbreed	Golden retriever	NS	NS	NS	Boerboel	Golden Retriever	Вохег
Sex	M	<u> </u>	ΙΉ	FN	FN	Σ	Σ	ΙΉ	M	M	NS	Ĭ L	Ÿ	MN
Age	11	2	∞	10	n/a	7	14	4	NS	NS	NS	9	10	∞
Author	Bindseil et al. [14]	Brown et al. [13]	Brown et al. [13]	Brown et al. [13]	Brown et al. [13]	Brown et al. [13]	Brown et al. [13]	Brown et al. [13]	Spangler and Culbertson [6]	Spangler and Culbertson [6]	Spangler and Culbertson [6]	Kitshoff et al. [4]	Clark et al. [8]	Rodinő Tilve et al. [7]
Case	1	2	m	4	ις	9	7	∞	6	10	11	12	13	41

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R. S	Diagnosis in authors opinion et al	Stalolipoma
	Original diagnosis Diagnosis in authors opini	Sialolipoma Sia
	Histological findings	Multilobular, encapsulated neoplastic mass composed of approximately 60% adipose tissue that entrapped normalappearing salivary tissue with a scant to moderate fibrovascular supporting stroma. Normal ducts were present within islands of salivary tissue with occasional abnormal ducts lined by large epithelial cells with voluminous, eosinophilic cytoplasm (oncocytic metaplasia) Frequently abnormal ducts were dilated with occasional intraductal mineralised material and surrounded by mild lymphocytic inflammation. The mass was surrounded by a thin, fibrous capsule that bordered adjacent non-neoplastic connective and adipose tissue.
	Follow up	No recurrence after 16 months
	Treatment	Excision
	Size	NS
	Duration of swelling/ clinical signs	4 weeks
	Salivary gland affected Duration of swelling/	Right parotid
	Age Sex Breed	FN Lurcher
	Age S	6
able 1 (continued)	Case Author	Rodinō Tilve et al. [7]
able	Case	15

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