

CASE REPORT

Companion or pet animals

An incidental finding of numerous hypocellular nodules within a mandibular–sublingual gland complex sialocoele in a dog

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Abstract

An 11-month-old female Labrador Retriever presented with a 3-month history of a progressively enlarging, unilateral, ventral mandibular swelling. Cytology was consistent with saliva, and the dog was referred for further investigations. Computed tomographic (CT) imaging was performed, and findings were suggestive of a left-sided mandibular sialocoele. The dog underwent sialoadenectomy surgery of the left mandibular–sublingual gland complex. On incising the thick capsule of the sialocoele, the swelling was incidentally found to contain hundreds of 1.5–2 mm firm, spherical structures. Cytological and histological examination of the structures demonstrated that these were non-mineralised, hypocellular, proteinaceous nodules. The nodules were not able to be identified on review of the pre-operative CT scan. To the authors' knowledge, this is the first report of such radiolucent, proteinaceous nodules being present within a canine sialocoele.

BACKGROUND

Sialocoeles are the most commonly presenting disease of the salivary glands of dogs.¹ This case report describes a case of a canine sublingual-mandibular sialocoele, with concurrent radiolucent, proteinaceous, hypocellular, nodules present within. Computed tomographic (CT) imaging has been considered the most sensitive imaging modality for diagnosing and differentiating salivary gland disease,² although sialolithiasis may be diagnosed by plain radiography due to the mineral component found in the sialoliths.^{2–5} In the presented case, the numerous nodules were not visible on the CT scan and found incidentally at surgery. Cytological and histological analyses revealed no mineral component to the nodules. To the authors' knowledge, this is the first case report describing the presence of radiolucent, non-mineralised nodules found within a canine sialocoele.

CASE PRESENTATION

An 11-month-old, female entire, Labrador Retriever presented to the referral hospital with a 3-month history of a progressive, soft, non-painful swelling in the left ventral mandibular region. The patient had no other significant medical history and was noted to be fed a raw diet. No trauma or known inciting cause had occurred prior to the owner noticing the mass, and the patient had no other associated clinical signs

such as pain or dysphagia. Cytological evaluation of aspirated fluid prior to referral demonstrated a small number of non-degenerate neutrophils and a high number of vacuolated and foamy macrophages in a pink mucinous granular background consistent with saliva. Many macrophages contained small amounts of haemosiderin, and erythrophagia was occasionally found, indicating chronic active haemorrhage. These findings were consistent with a sialocoele.

INVESTIGATIONS

On presentation, the clinical examination confirmed the presence of a 6 × 3.5 cm firm, discrete swelling in the left ventral mandibular region. CT images of the head and neck were acquired under sedation by use of a 64-row multidetector CT-scanner (Somatom Definition AS Siemens, Erlangen, Germany). Images were obtained pre- and postapplication of iodinated non-ionic contrast medium (Iopamidol-Niopam 350, Bracco UK, High Wycombe, UK) injected intravenously at a dose of 2 ml/kg. The injection of contrast medium was followed by the injection of 10 ml of saline solution.

CT images revealed a 6.2 × 3.4 cm, well-defined, non-contrast enhancing, homogenous, cystic mass (mean 16.1 Hounsfield units, consistent with fluid), with a thin contrast enhancing rim. The mass had an extensive surface of intimate contact with the left mandibular salivary gland, which appeared compressed and mildly displaced medially and

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caudally (Figure 1). No sialoliths were noted on the scan. The surrounding tissues were unremarkable, with no evidence of cellulitis or subcutaneous oedema. The left medial retropharyngeal lymph node was mildly enlarged (1.3 cm), and the radiographical diagnosis was consistent with a left-sided mandibular–sublingual complex sialocoele with most likely reactive left medial retropharyngeal lymphadenopathy.

TREATMENT

Surgery was performed 24 h after the CT scan, under general anaesthesia. The dog was premedicated with 0.3 mg/kg methadone (Comfortan, Dechra) and 2 µg/kg dexmedetomidine (Dexdomitor, Zoetis) intravenously, and then induced with propofol (PropoFlo, Abbot) to effect. The dog was intubated and maintained on isoflurane and oxygen. A left ventral paramedian surgical approach was made to access the sialocoele and associated salivary glands. On incising the thick capsule of the sialocoele, hundreds of small 1.5–2 mm firm, spherical structures within viscous haemorrhagic fluid were released from the swelling (Figure 2a,b). A question was initially raised over there being parasitic involvement, especially since the dog was fed a raw diet. As a result, a sample of the fluid with some spherical structures was submitted for rapid laboratory analysis to rule out zoonotic risk.

The spherical structures were observed under the microscope, and a squash preparation was obtained and stained with May–Grünwald–Giemsa stain. The material was dense and hard, and it consisted of indistinct cells, extracellular pink matrix, and haematoidin crystals, suggesting a proteinaceous nodule (Figure 3a,b). Cytological examination of the fluid demonstrated a large amount of viscous, eosinophilic to basophilic, amorphous material (saliva and mucous), a large amount of blood, several foamy macrophages and a few haematoidin crystals (Figure 3c,d). This was consistent with the previous diagnosis of sialocoele with chronic haemorrhage.

Surgery was continued, and a routine left mandibular and sublingual sialoadenectomy was performed. The duct was ligated as rostrally as possible at the level of the lingual nerve and transected. A portion of the sialocoele capsule, a few nodules

LEARNING POINTS/TAKE-HOME MESSAGES

- Sialocoeles may contain proteinaceous nodules in addition to saliva.
- Nodules may not be readily visible with preoperative imaging and may be an incidental finding at surgery.
- The presence of numerous nodules within a sialocoele should not be confused with parasitic cystic infestation.

and the affected mandibular and sublingual salivary glands were submitted for further histopathological analysis.

OUTCOME AND FOLLOW-UP

The patient recovered well from surgery and was discharged from the hospital 2 days postoperatively with oral paracetamol (Paracetamol, Summit) at 10 mg/kg twice daily, and meloxicam (Loxicom, Norbrook) at 0.1 mg/kg once daily. The dog developed a seroma within the 24 hours following surgery, but this resolved without further treatment.

Further analysis of sections of the nodules was achieved after embedding them in agarose (Noble agar, cat. No. A543, Sigma-Aldrich), followed by routine histological processing and staining with haematoxylin and eosin (H&E), Masson's trichrome (MT), Von Kossa and periodic acid Schiff (PAS)-Alcian blue. On cross-section, the spheres consisted of hypocellular, proteinaceous material with a subtle suggestion of concentric pattern to fibroblasts scattered throughout (Figure 4). No mineralised material was detected on H&E or Von Kossa's stains. The nodules stained diffusely blue with MT, indicating a prevalent collagenous component, and were strongly PAS and weakly Alcian blue positive, indicating a marked mucinous component. Histologic examination of the sialocoele capsule revealed a fibrous wall markedly expanded with oedema. The internal surface was segmentally covered by fibrin, extravasated erythrocytes and neutrophils. The

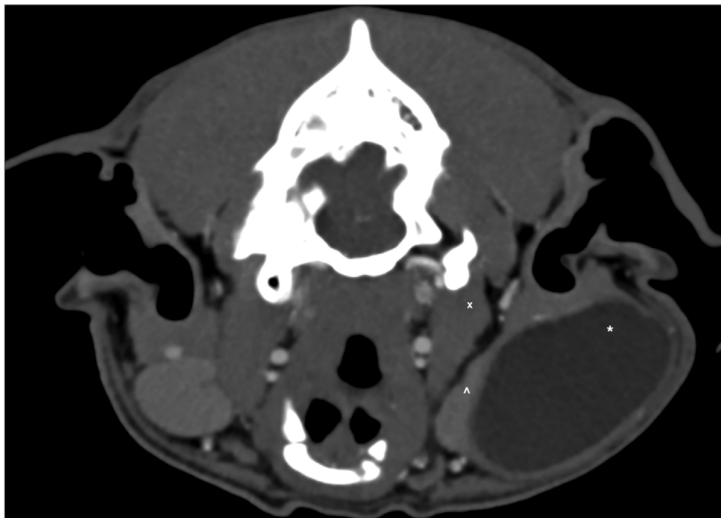


FIGURE 1 Transverse three-dimensional CT image reconstruction of the head at the level of the cystic lesion, sialocoele (*), left mandibular salivary gland (^) and the digastric muscle (x̄).

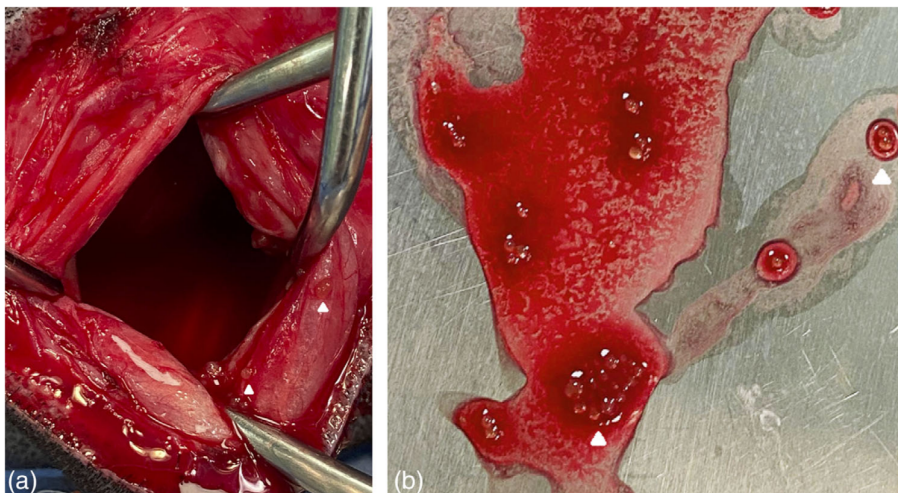


FIGURE 2 (a) Intraoperative image showing cystic/sialocoe capsule and multiple nodules (white arrowhead) around the periphery of the surgical site. (b) Image showing multiple glistening, spherical nodules within haemorrhagic fluid intraoperatively

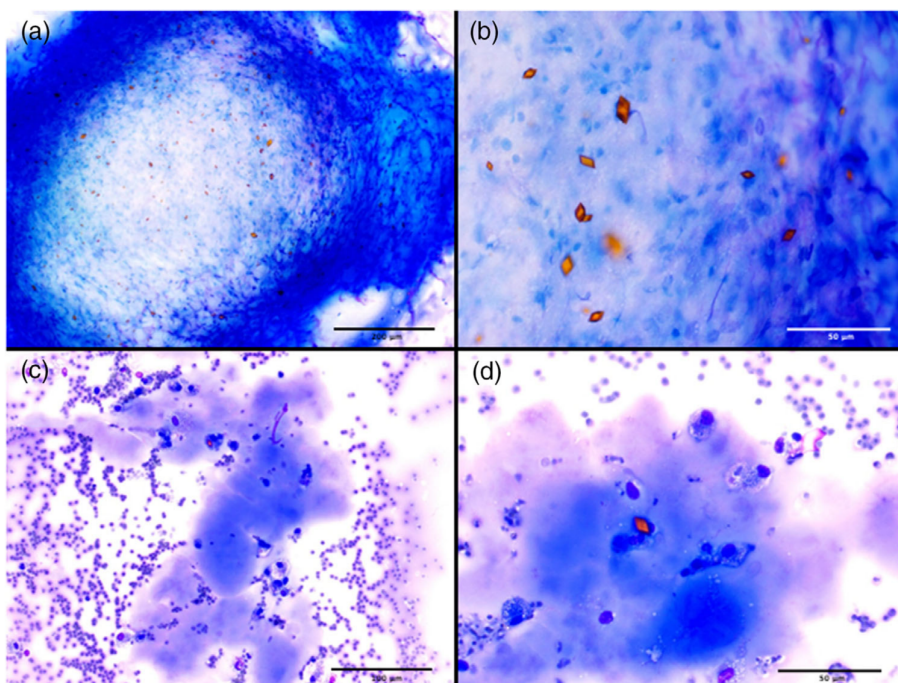


FIGURE 3 (a and b) Squash preparation of a nodule from the sialocoele, demonstrating the presence of indistinct cells admixed with extracellular pink matrix, and haematoidin crystals, May-Grünwald-Giemsa stain: (a) 10X objective, bar 200 μm , (b) 40X objective, bar 50 μm . (c and d) Sialocoele cytology with lakes of amorphous basophilic to eosinophilic extracellular material (saliva and mucous) associated with foamy macrophages and haematoidin crystals: (c) 10X objective, bar 200 μm ; (d) 40X objective, bar 50 μm

deep layer of the wall contained scattered haemosiderin-laden macrophages. In the superficial layer of the wall, occasional round collections of hypocellular, eosinophilic, proteinaceous material were found.

At 6 months after surgery, the dog was clinically well and had no recurrence of the sialocoele.

DISCUSSION

Sialocoeles, otherwise known as salivary mucocoeles, are the most commonly reported disease of the canine sali-

vary glands. A sialocoele is a collection of saliva in the subcutaneous tissues that becomes encapsulated due to the inflammatory reaction caused by the extravasation of saliva.^{5,6} They most likely form due to damage to the salivary gland or one of its ducts but the pathogenesis remains unclear. Trauma, foreign bodies, sialolithiasis and neoplasia have all been cited as possible causes.^{7,8} The cause in this case is not known.

Sialolithiasis accounts for approximately half of salivary gland disease in humans and describes the presence of a calculus or calculi within a salivary gland.⁹ Sialolithiasis in dogs is reported infrequently, but it has been reported to affect all of the major salivary glands.^{2,5,10} The composition

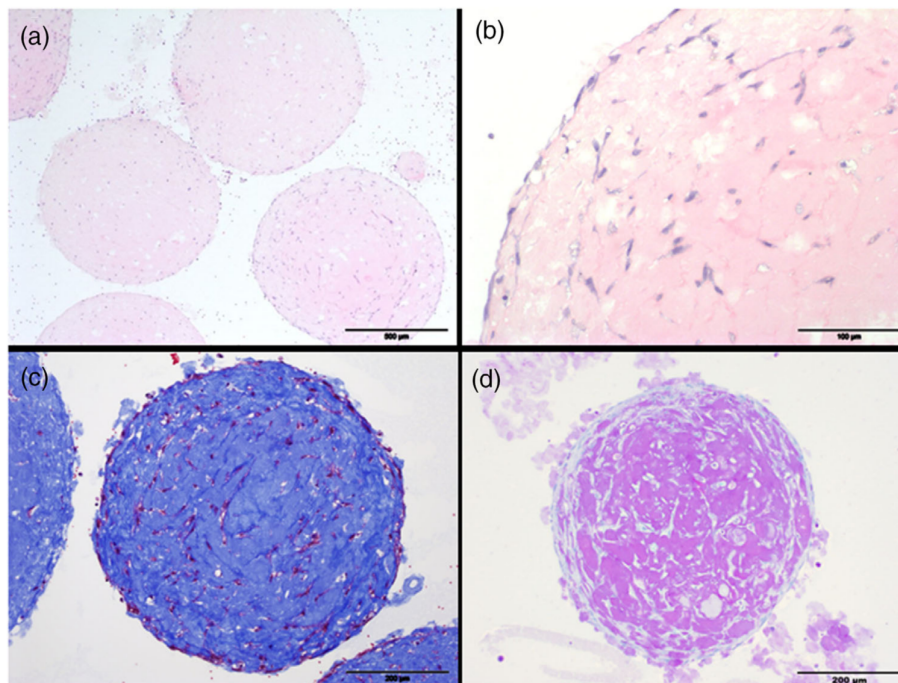


FIGURE 4 Nodule from the sialocoele of an 11-month-old dog consisting of primarily proteinaceous matrix embedded with low numbers of fibroblasts. (a) Haematoxylin and eosin stain 4× objective, bar 500 μm ; (b) haematoxylin and eosin stain 20× objective, bar 100 μm ; (c) Masson's trichrome stain 10× objective, bar 200 μm ; (d) combined periodic acid Schiff-Alcian blue stain 10× objective, bar 200 μm

of canine sialoliths is variable, but by definition, the majority are reported to have some inorganic mineral component. Most commonly these stones have a calcium salt component (oxalate, carbonate, phosphate) and sometimes a magnesium component.^{2,3,5,10–12} Reports in human medicine suggest that the majority of sialoliths also have a mineral component. In one human study analyzing sialoliths from 21 different patients, 18 of 21 had a mineral component, while three of 21 contained only organic material.⁹

There has only been one veterinary case report of nodules in a canine salivary gland that lacked a mineral component, reported to be 100% proteinaceous material, yet the nodules were still easily visible as radiodense structures on plain radiographs.⁴ If, as in our case, the nodules lacked a mineral component, then arguably the term sialolith may have been misplaced. Osseous metaplasia has been reported within canine sialocoeles,^{13,14} and this could have provided an explanation for the radiodense appearance of the salivary glands in the previous case. In our case, the nodules lacked a mineral component on cytological and histological analyses and were found to be made up of a combination of collagenous and mucinous material, haematoidin crystals and cellular debris, making these purely organic nodules.¹⁵ Due to the presence of secretory mucoid material within the nodules, we hypothesise that these nodules were formed by means of fibroblasts organising inspissated secretory material. Because mucocoeles are hollow structures, multilocular botryoid proliferation into the luminal space is a logical pattern of fibrovascular proliferation and would result in spherical inclusions within the mucocoele wall and lumen. Similar botryoid proliferation has been described in canine and human rhabdomyosarcomas.^{16,17} It remains unclear why they were present in such a large number in this case.

Different imaging modalities can be useful in diagnosing and differentiating salivary gland disease. In human medicine, CT is considered the modality of choice for diagnosing sialolithiasis and has a superior sensitivity to other modalities.¹⁸ It is also considered the most effective way to diagnose the specific gland of origin for a sialocoele, and to look for involvement of nearby structures and allow for effective surgical planning.¹ In this case, the nodules were not visible on the CT scan, which to the authors' knowledge is not yet something that has been previously reported.

Surgical treatment of sialocoeles is recommended, due to high recurrence rates with medical management alone.⁶ Sialoadenectomy is recommended due to high recurrence rates with drainage or marsupialisation of the sialocoele without gland removal.⁶ A ventral paramedian approach was chosen due to surgeon preference and is supported by a recent paper, suggesting a lower recurrence rate than using a lateral approach.¹⁹ There are reports of sialolith removal from the parotid duct, in cases of isolated sialolithiasis, which is then left to heal by second intention or surgically repaired.^{11,12} This option is likely to be less appropriate for cases with concurrent sialocoeles, ductal fibrosis or in the case of multiple nodules being present within a sialocoele. In our case, sialoadenectomy with concurrent removal of the contained nodules was the surgical treatment of choice.

In conclusion, this clinical case report describes the presence of a sialocoele containing large numbers of hypocellular, proteinaceous (fibromucinous) nodules in a young Labrador Retriever. A CT scan did not identify the nodules prior to surgery but surgical management was curative, with no signs of recurrence 6 months postoperatively.

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CONFLICTS OF INTEREST

The authors declare they have no conflicts of interest.

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The authors received no specific funding for this work.

ETHICS STATEMENT

As a client-owned animal, treated as a normal clinical case, no ethical approval was required for this retrospective case report.

AUTHOR CONTRIBUTIONS

All authors have meaningfully contributed to the production and editing of this manuscript and have reviewed the final version.

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