

## Minor salivary gland sialolithiasis associated with a palatal sialocele in a dog

### *Sialolithiasis in een palatale sialocele bij een hond*

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## ABSTRACT

A five-year-old Labrador retriever was referred for progressively worsening inspiratory stridor and accompanying dyspnea. Oropharyngeal inspection identified a mass in the soft palate as the cause for upper airway obstruction. Fine-needle aspiration of the mass evacuated mucohemorrhagic fluid and revealed multiple sialoliths. The sialocele was surgically explored, all sialoliths were removed, the lining of the sialocele was resected, and the remaining defect in the palate was reconstructed and closed. Histology of submitted tissues confirmed it to be a sialocele and the sialoliths consisted of protein and carbonate apatite. The dog made an unremarkable recovery and was discharged on the same day. No recurrence occurred and the dog was free of any respiratory symptoms six months post treatment.

## SAMENVATTING

Een vijf jaar oude labrador-retriever werd aangeboden met progressief verergerende inspiratoire stridor en dyspnee. Bij orofaryngeaal onderzoek werd een massa in het zachte gehemelte geïdentificeerd als de oorzaak van de bovensteluchtwegobstructie. Via aspiratie van de massa werd een mucohemorragische vloeistof gedraineerd met daarin verschillende steentjes. Er werd een sialocele vermoed die chirurgisch werd geëxploreerd. Alle sialolieten en de epitheliale aflijning van de massa werden verwijderd. Het defect in het palatum werd nadien gereconstrueerd. Histologisch onderzoek bevestigde dat het een sialocele betrof met daarin sialolieten bestaande uit eiwit en carbonaat. De hond herstelde zonder complicaties en werd dezelfde dag uit het ziekenhuis ontslagen. Zes maanden na de ingreep was de hond nog steeds vrij van respiratoire klachten.

## INTRODUCTION

Dogs have four pairs of major salivary glands, sometimes also referred to as the extra-oral salivary glands (parotid, mandibular, sublingual, and zygomatic) (Gil et al., 2018). Additionally, numerous minor salivary glands are distributed throughout the oral cavity, such as the lips, cheek, tongue, palate, pharynx, and esophagus. Salivary gland disease is uncommon with an incidence of 0.3% (Furtado et al., 2017). Most reported diseases concern the major salivary glands, including nonspecific sialadenitis, neoplasia, sialocele, sialolithiasis, salivary gland lipomatosis, necrotizing sialometaplasia, and traumatic hemor-

rhage (Spangler and Culbertson, 1991; Lieske and Rissi, 2020).

Sialolithiasis, the formation of stones in saliva, is a rare condition in dogs, mostly associated with the parotid salivary gland (Jeffreys et al., 1996; Trumpatori et al., 2007; Proot et al., 2013), the mandibular salivary gland (Pignone et al., 2009; Shuh et al., 2015), the sublingual salivary gland (Ryan et al., 2008), and the zygomatic gland (Lee et al., 2014). The sialolith formation can be caused by mineral aggregation in the salivary gland or duct together with the accumulation of mucus, peel epithelial cells, bacteria, and the presence of an alkaline pH (Pignone et al., 2009). In a study by Schroder et al. (2017), increased concentra-

tions of the ions that constitute the inorganic phase of sialoliths were found when the saliva of dogs with sialolithiasis was compared to control dogs (Schroder et al., 2017). But more often, sialoliths occur concurrently with a sialocele, a collection of mucoid saliva that has leaked from a damaged salivary gland or duct (Ryan et al., 2008; Torad and Hassan, 2013). Stone formation has been attributed to the sluggish salivary flow within a chronic sialocele and the potential increase in the number of bacteria via salivary retention (Han et al., 2016).

In the current case, the presence of a palatal sialocele containing multiple sialoliths is described. To the best of the authors' knowledge, this is the first description of sialolithiasis associated with a minor salivary gland in a dog.

### CASE HISTORY

A five-year-old, male, entire Labrador retriever weighing 41 kg was referred to Anicura Dierenkliniek De Ark for progressive worsening of inspiratory stridor and exercise intolerance over the last year.

Before the development of the clinical symptoms, the dog was very active and known to retrieve and play with wooden sticks. Over a period of around four months, he progressively developed an inspiratory stridor, resulting in exercise intolerance and periods of dyspnea. There was no dysphagia. On presentation, clinical examination was unremarkable apart from a stridor. The dog did not have enlarged submandibular lymph nodes, opening of the mouth was not painful, body temperature was 38.5°C, neurological examination and thoracic auscultation were normal. The stridor

consisted of a combined pharyngeal and laryngeal component. When the pharyngeal region was transcutaneously palpated, the dog became uncomfortable, had an obvious swallowing reflex, and showed worsening signs of stridor.

Because of the breed's predisposition for laryngeal paralysis, laryngeal inspection was performed as the next diagnostic step. The dog was intravenously (IV) premedicated with 0.2 mg/kg methadon (Insistor, Eucuphar, Belgium) and 5 µg/kg dexmedetomidine (Dexdomitor, Orion pharma, Belgium) and maintained with boli of 2 mg/kg alfaxalone (Alfaxan, Eurox, the Netherlands). The dog also received 0.15 mg/kg meloxicam (Cticam, Eucuphar, Belgium). To allow a patent airway during the examination, a 9-mm single-use endotracheal tube (Endotracheal tube cuffed straight, Teleflex medical, Ireland) was introduced in the dog's trachea and the cuff was inflated.

A marked asymmetrical swelling was noted at the soft palate (Figure 1). The mass measured approximately 6 x 3 x 3 cm and was located just medial to the right tonsil. Due to its mass effect, the right tonsil was everted from its crypt. On digital palpation, the mass had both fluctuating and dense regions. Further inspection of the mouth and laryngeal region showed no other abnormalities. Aspirates were taken and 20 ml of a mucohemorrhagic fluid was drained. Inside the fluid, some miniscule sialoliths were present and while performing the fine-needle aspiration, larger mineralized structures were felt (Figure 2).

The dog was transferred to the surgical room and positioned in sternal recumbency with his head fixed to a frame with the mouth open (Findji and Dupré, 2008). He received 20 mg/kg cephalosporin (Cefazoline, Sandoz, Belgium) IV. The oropharyngeal mucosa

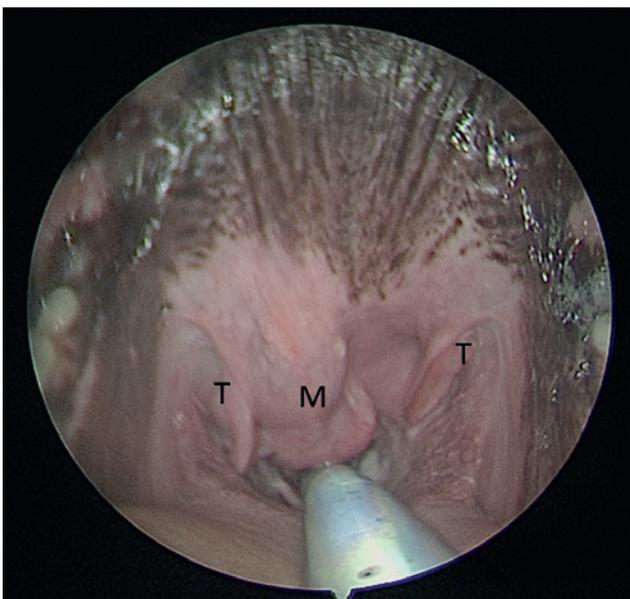


Figure 1. Endoscopic view of the pharynx demonstrating bilateral everted tonsils (T) and the asymmetrical bilobed swelling of the soft palate mucocoele (M) responsible for upper respiratory obstruction.

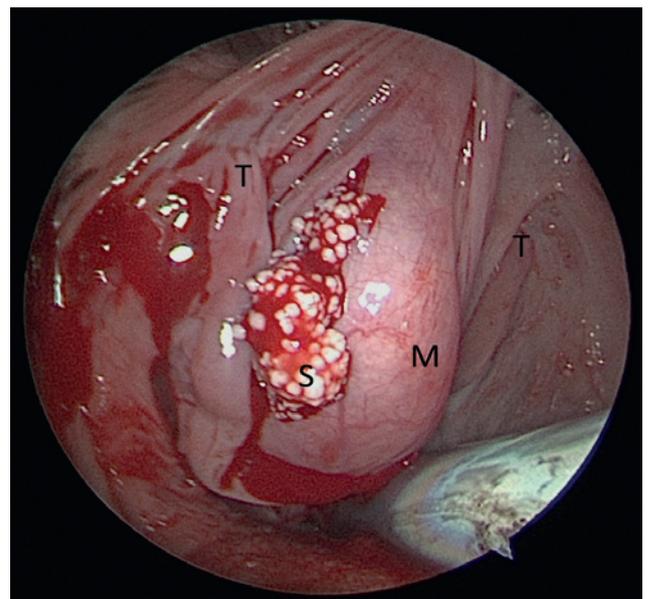


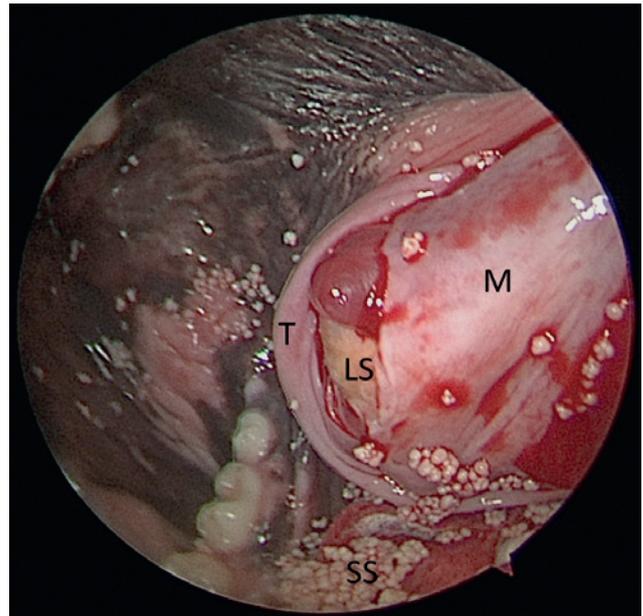
Figure 2. Endoscopic close-up view of the mass after drainage of 20 ml of mucohemorrhagic fluid revealed multiple small sialoliths exiting the puncture hole (T: tonsil, M: mucocoele, S: sialolith).

overlying the mass was incised with a nr 15-scalpel blade revealing a bilobed, fibrous capsule containing one large sialolith (6 mm diameter) surrounded by thickened mucus in one pocket, and numerous small sialoliths (1-2 mm diameter) in the second pocket (Figure 3). All sialoliths were evacuated, and the fibrous capsule was removed en-bloc with the adjacent right tonsil (Figure 4). The resulting palatal defect was closed in two continuous appositional layers with a 4/0 slow-resorbable monofilament suture (Maxon, Covidien, France).

The dog could be extubated without complications. He was discharged the same day with a five-day-oral course of 15 mg/kg cephalosporin (Cefaseptin, Vetoquinol, Belgium) twice daily and a five-day-oral course of 2 mg/kg cimicoxib (Cimalgex, Vetoquinol, Belgium) once daily.

Bacteriological culture of the stagnant saliva was negative. Histologic analysis of the tissue identified a pseudocystic wall of granulation tissue and neovascularization with mixed inflammation oriented around accumulations of fibrin, necrotic cellular debris, mineralized debris, and mucoid material. Some of the surrounding salivary ducts were dilated and contained inspissated mucus. There were foci of lymphonodular hyperplasia in the right tonsil. Quantitative analysis showed that the sialoliths were composed of 80% protein and 20% carbonate apatite (Figure 5).

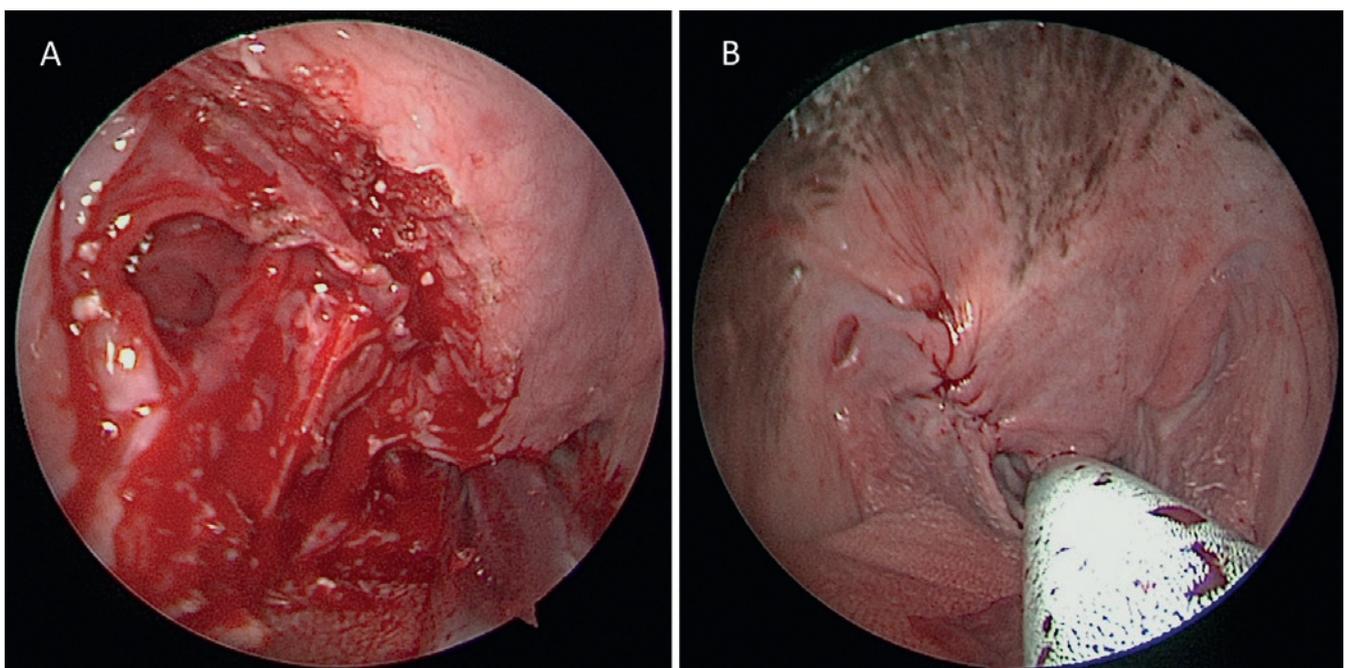
Telephone check-ups were made at one day, ten days and finally six months post-operative. No complications occurred during this period and the owner reported the dog to be active again, without any respiratory signs.



**Figure 3.** Intraoperative view after incision of the palatal mucosa to open the sialocele demonstrating the presence of a large sialolith inside (T: tonsil, M: mucocoele, LS: large sialolith, SS: small sialolith).

## DISCUSSION

Causes for pharyngeal/laryngeal stridor in the dog include brachycephalic obstructive airway syndrome, laryngeal paralysis, pharyngeal stick injury, neoplasia (lingual, tonsillar, laryngeal), the presence of a foreign body, and hyoid trauma. Respiratory obstruction and distress due to salivary gland disease is most often caused by the presence of a pharyngeal mucocoele af-



**Figure 4.** Intraoperative view of the palatal defect. A. After resection of the sialocele and right tonsil and B. after reconstruction and two-layer closure.



**Figure 5.** Close-up view of the carbonate apatite sialolith (picture taken with sialoliths in a plastic recipient).

ter trauma to the sublingual and/or mandibular glands or ducts (Furtado et al., 2017). Clinical signs of salivary gland disease, in case of obstruction of ducts by sialoliths, include painful or non-painful (subcutaneous) swelling surrounding the affected glands (Suh et al., 2015; Yoon et al., 2017). In a case report by Han et al. (2016), a thirteen-year-old, mixed-breed dog with a pharyngeal sialoceles containing sialoliths that were responsible for respiratory signs such as intermittent coughing, and choking was described. But also swelling of the soft palate due to minor salivary gland disease will cause upper airway obstruction and dyspnea. This has been described by Watanabe et al. (2012), when a sialoceles was identified originating from a minor salivary gland in the soft palate of a four-year-old Welsh corgi (Watanabe et al., 2012). Wainberg et al. (2020) reported upper airway obstruction due to a mucus retention cyst in the soft palate of a ten-year-old Chihuahua. The authors referred to the pathology as a mucus retention cyst rather than a mucocele, since its outer layer consisted of an uninterrupted epithelial layer instead of mostly consisting of granulation tissue.

If sialoliths are located in the excretory duct of major salivary glands, the sialoliths can sometimes be removed using duct incision via a transcutaneous or intraoral approach (Han et al., 2016). For the surgical treatment of a sialoceles, there are several options. Major salivary gland duct rupture can be solved by transposition of the duct to a new location, when this is anatomically feasible (Yoon et al., 2017). Repair by end-to-end anastomosis over a stent is another option, but often not possible because of duct fibrosis (Trumpatori et al., 2007). Ligating a traumatized duct proximal to its severed ends leads to glandular secretory cell atrophy because of back pressure (Yoon et al., 2017). Finally, the major salivary gland can be removed together with its draining duct (Suh et al., 2015; Proot et al., 2016; Han et al., 2016).

The only described dog with a palatal sialoceles was treated for almost two years by repeated puncture and drainage, before marsupialization was performed, and finally marginal resection led to resolution of the

sialoceles (Watanabe et al., 2012). In this case, conservative treatment (drainage and marsupialization) could only partially resolve the space-occupying effect and would likely have resulted in recurrence of potentially dangerous respiratory obstruction. Immediate surgical resection of the sialoceles was therefore performed. In major salivary gland disease, both sialolith removal and sialoadenectomy lead to good outcomes and low recurrence rate, based on a small number of cases (Proot et al., 2016; Han et al., 2016). The dog with the mucus retention cyst also underwent immediate surgical resection after diagnosis, without any attempt at conservative treatment, and had no recurrence after a seven-month follow-up (Wainberg et al., 2020).

Salivary gland diseases are more likely to occur in middle-to-old aged dogs (Spangler and Culbertson, 1991; Han et al., 2016). In young dogs, salivary gland disease has been attributed to the presence of a migrating foreign body in the parotid duct of a fourteen-month-old boxer (Marques et al., 2008), oral trauma with rupture of the excretory ducts of a zygomatic salivary gland in a two-year-old Australian shepherd (List and Goldhammer, 2012), and a bite wound to the neck of a three-year-old French bulldog (Yoon et al., 2017). Given the history of playing with wooden sticks, the authors assume that a traumatic origin is also likely in this case. Trauma to the oropharyngeal palatal mucosa and the ensuing healing may lead to fibrosis of excretory ducts resulting in the accumulation of saliva in the soft palate.

A diagnosis of sialolithiasis can be made by palpation, radiography, ultrasonography, or sialography (Lee et al., 2014; Suh et al., 2015; Proot et al., 2016). Diagnosis of major salivary gland mucoceles is routinely performed by ultrasonographic examination (Torad and Hassan, 2013). For the palatal sialoceles, in this case, ultrasonography was not an option. Plain radiographs on the other hand, could have been helpful. They have been used to differentiate sialoceles from bony lesions, aggressive neoplasms of the mandibular region, or diagnose sialolithiasis (Torad and Hassan, 2013). Even non-mineral sialoliths can be radiopaque and visible on plain radiographs (Suh et al., 2015). Advanced medical imaging (computed tomography and magnetic resonance imaging) has also been described for the major salivary glands in dogs (Weidner et al., 2012; Lee et al., 2014; Durand et al., 2016; Gil et al., 2018). In human medicine, magnetic resonance imaging (MRI) of the salivary glands is typically used to assess the extension and infiltration of tumors, particularly into the parapharyngeal space (Weidner et al., 2012). CT sialography has been successfully described for the parotid, zygomatic and mandibular salivary gland in dog cadavers (Kneissl et al., 2011).

Analysis of sialoliths in veterinary medicine has demonstrated calcium carbonate, calcium phosphate, magnesium carbonate, and magnesium ammonium phosphate (Trumpatori et al., 2007; Han et al., 2016).

In the current case, only a 20% mineral content was found and 80% of the sialoliths consisted of a non-mineral proteinaceous material. Non-mineral sialoliths have also been found by Suh et al. (2015) who described a ten-year-old Maltese dog with bilateral radiopaque sialoliths in the mandibular gland that consisted completely of proteinaceous material. They hypothesized that the highly condensed nature of the sialoliths accounted for their radiopaque nature. This could also explain the scratching sensation that was felt when performing fine-needle aspiration of the mucocele in the current case. The mineralization in this case most likely consisted of secondary mineralization around condensed proteinaceous material, but also ectopic ossification presenting as osteoid metaplasia in a sialocele has been described in a four-year-old Shih Tzu (Fernandes et al., 2012).

## CONCLUSION

Accumulation of saliva in a palatal mucocele with the concurrent development of palatal sialoliths is a rare differential diagnosis for respiratory obstruction. Surgical removal of the sialoliths and the accompanying sialocele is curative.

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