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Surgical Removal of a Cervical Sialocele in a 9-Year-Old Intact Female Shih Tzu

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Abstract

Background: A sialocele is an accumulation of saliva in the subcutaneous tissues after leakage from a damaged salivary gland or duct. A 9-yearold, intact, female, Shih Tzu was brought to the UP Veterinary Teaching Hospital and presented with a firm mass on the left submandibular to cranial cervical area. Based on the patient history, physical examination, diagnostic imaging, and cytology, the dog was diagnosed with a cervical salivary mucocele or sialocele, possibly due to a defect in the left mandibular to sublingual salivary glands or gland-duct complex. A prior treatment regimen using corticosteroids and antibiotics had no effect on the size and consistency of the sialocele, further indicating surgical removal. Methodology: Surgical management via lateral approach was done to treat the sialocele via excision of the left mandibular and sublingual salivary glands along with their associated salivary ducts. Results: Treatment resulted in good post-operative healing in the submandibular to cervical region with no observed recurrence of sialocele. Conclusion: Histopathologic findings of the excised mass confirmed the sialocele and revealed focally extensive granulation tissue, multiple sialoliths, and sialadenitis. Case findings support that lateral approach

sialoadenectomy was effective treatment for the patient's ailment.

Keywords

Keywords: canine, cervical, sublingual gland, sialocele, sialoadenectomy

1. Introduction

A salivary mucocele or sialocele is the accumulation of saliva in the subcutaneous tissue due to leakage from a damaged salivary gland or salivary duct [1, 2]. It differs from a cyst since it lacks luminal epithelium and is instead lined by granulation tissue, which arises from the inflammatory reaction to free saliva [2]. There are four different types of sialoceles based on location: cervical sialoceles form in the intermandibular area, sublingual sialoceles under the tongue, pharyngeal sialoceles within the throat, and zygomatic sialoceles just below the eye. The sublingual gland and duct complex are the structures most associated with a sialocele [3, 4, 5]. In dogs, sialoceles occur more commonly in the cervical and sublingual regions [6]. Sialoceles originating from the mandibular and sublingual salivary glands can also affect dogs of various breeds and in differing age groups [7]. The location of the sialocele will often determine the presenting complaint

and indicate which salivary gland/s it arises from. Most dogs with sialoceles present with a history of ventral mandibular or cranio-ventral swelling in the neck, classifying it as a cervical mucocele or cervical sialocele [8], as seen in this case. Cervical sialoceles present as gradually developing neck masses that are soft, fluctuant, and either minimally sensitive or non-painful. Patients exhibit pain usually only during the acute phase of the sialocele [1]. Surgical removal of the mass, which consists of saliva, inflammatory tissue, and the involved salivary glands, is an effective treatment. Recurrence or serious complications are rarely reported [9, 10].

Earlier research indicates that sialoceles are underdiagnosed and seldom documented in dogs. Spangler and Culbertson [11] reported only a 9% prevalence of sialoceles from diagnostic pathology records from July 1985 to November 1988. Shivaraju *et al.* [12] report the occurrence as less than 20 out of 400 dogs. Medical records obtained from the University of the Philippines Veterinary Teaching Hospital – Diliman Station [13], dated 2011-2021, revealed an average of 3 cases of sialoceles per year, most being cervical in origin. This coincides with the 0.3% prevalence rate of salivary gland disease in dogs and cats according to Soltero-Rivera and Reiter [6]. To the knowledge of both authors, this is the first reported Philippine case of surgical removal of the sublingual and mandibular salivary glands for the treatment and management of a cervical sialocele in a canine patient. This case report

aims to describe the clinical presentation, diagnosis, and successful surgical excision of affected salivary glands, in that it may serve as a future reference in the treatment of similar salivary mucoceles.

2. Case Presentation

A 9-year-old intact female Shih Tzu with swelling in the left cranial cervical region was brought to the UP Veterinary Teaching Hospital – Diliman station, to obtain a second opinion. The swelling was first observed 6 months before presentation. A firm, irregularly shaped, and non-painful mass was palpated in the swollen area of the neck. No dysphagia or dyspnea was caused by the swelling in the neck. None of the associated lymph nodes were inflamed and no anatomical abnormalities or clinical signs of systemic illness were observed.

The use of corticosteroids and antibiotics did not soften or shrink the mass. The mass had well-defined borders and measured approximately 4.5cm x 3.0 cm xf.0 cm. Body temperature, heart rate, and respiratory rate of the dog were all within normal range. The complete blood count (CBC) showed no abnormal values except for a minute decrease in packed cell volume (PCV). Blood chemistry values were all within the reference range except for slightly elevated globulin (Table 1). No significant findings could be gathered from the blood test results.

Table 1. Complete blood count and blood chemistry results of the patient. The blood test results show no significant findings. CBC was done using a Mythic® 18 veterinary hematology analyzer and blood chemistry was read using a MNCHIP® dry chemistry analyzer. Reference values: Plumb, D.C. [14]v

Parameters	Patient Values	Reference Range		
Total white blood cell (x10 ³ /uL)	16.1	4.0-18.2		
Segmented neutrophil (%)	13, 685 (85)	2500-15700		
Lymphocytes (%)	1,610 (10)	300-3900		
Eosinophils (%)	644 (04)	0-1300		
Monocyte (%)	161 (1)	0-1,400		
Total red blood cell (x106/uL)	5.32	4.48-8.53		
Hemoglobin (g/dL)	13.7	10.5-20.1		
Packed Cell Volume/PCV (%)	32.5	33.0-58.7		
Platelet (x10 ³ /uL)	318	140-540		
Mean Corpuscular Volume (fl)	55.6	63-78.3		
Mean Corpuscular Hemoglobin	42.0	21.0-27.0		
(pg)	42.0	21.0-27.0		
Mean Corpuscular Hemoglobin	23.4 30.8-35.9		23.4	30.8-35.9
Concentration (f/dL)	20.4	90.0-99.9		

2.6	2.6-4.0
7.3	5.0-8.3
4.7	2.2-4.1
0.6	<u> </u>
9.4	8.7-12.0
111	74-145
17.9	8.0-30.0
3.89	2.5-7.9
1842	268-1653
306	98-300
43	14-151
0.21	0.1-0.5
50	13-289
0.37	0.4-2.0
204	50-554
48	-
	7.3 4.7 0.6 9.4 111 17.9 3.89 1842 306 43 0.21 50 0.37 204

Reference values: Plumb, D.C. [14]

Radiographic evaluation showed a radio-opaque irregularly shaped mass in the subcutaneous layer ventral to the angular process of the caudal mandible (Fig. 1). Ultrasonography revealed a round hypo-echoic section with distinct borders and hyperechoic margins. This indicated fluid leakage in between layers of subcutaneous tissue (Fig. 2). A cervical salivary mucocele was suspected based on patient history and clinical findings. Further

evaluation was done via fine needle aspiration of the mass under aseptic conditions. A thick, white-colored mucoid fluid was aspirated from the mass. Microscopic examination of the aspirate showed the presence of inflammatory cells and salivary crystals (Fig. 3), confirming sialocele formation. Surgical management to excise the mass and the involved mandibular and sublingual salivary glands was selected as the treatment.





Fig. 1. Radiograph images of the cervical region of the dog patient. (A) Lateral view and (B) dorsoventral view show a subcutaneous cervical mass (red outline) located caudoventral to the angular process of the mandible. Images were processed using a Fujifilm® Skanmobile and PrimaT2 digital radiograph machine.



Fig. 2. Ultrasonographic image of the cervical mass. An anechoic region (yellow arrow) with a hyperechoic border (red circled) indicates a fluid-filled cavity surrounded by a well-defined thick border. Ultrasonography was performed using a Well.D $^{\circ}$ Veterinary Ultrasound Scanner.

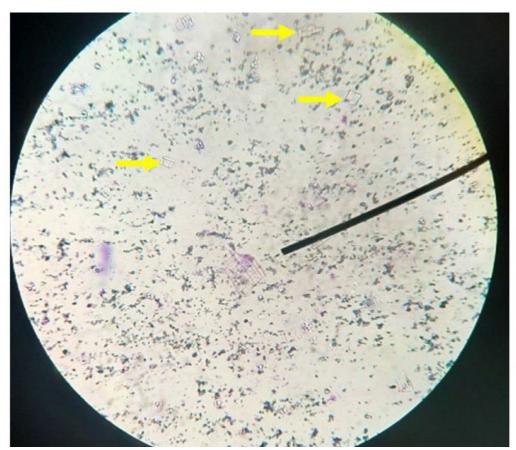


Fig. 3. Microscopy of aspirated fluid from the mass. Examination of the fluid showed the presence of salivary crystals (yellow arrows). Magnification 1000x/Examined under oil immersion objective. Diff Quik® staining was used.

3. Treatment

The cervical sialocele was surgically treated via sialoadenectomy. The goal of sialoadenectomy is to remove the defect where salivary leakage is occurring. To do so, the damaged mandibular and sublingual salivary glands and the affected gland duct complex are excised. Atropine sulphate (0.04 mg/kg SC) was injected pre-anesthesia to inhibit excessive salivary and bronchial secretions. Anesthesia was induced using the combination of zolazepam and tiletamine (5mg/kg IV). The dog was intubated, and the patient was maintained on gas anesthesia with isoflurane. The patient

was positioned on right lateral recumbency to allow lateral approach access to the left cervical region. The head and neck are placed on a soft cloth to prevent damaging the right side of the head during surgery. The skin incision was made over the region of the mandibular salivary gland vertically towards the ventral linguofacial vein (Fig. 4). After dissection through skin and subcutaneous tissues, the platysma muscle was incised from the angle of the mandible caudally to the external jugular vein. A firm, singular, irregularly shaped mass was bluntly dissected and exteriorized from the cranioventral cervical region (Fig. 5A).

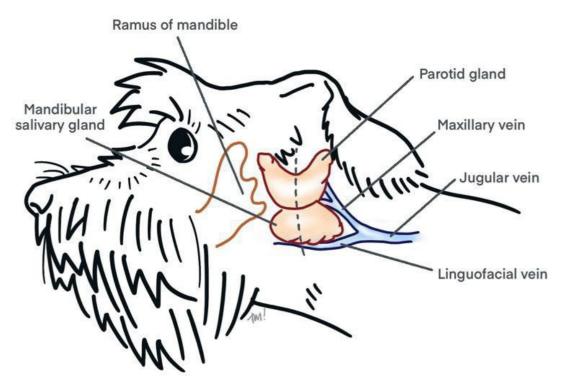


Fig. 4. Anatomic illustration for sialoadenectomy incision. A vertical incision (dotted line) is made over the mandibular salivary gland to access the affected salivary glands and sialocele.

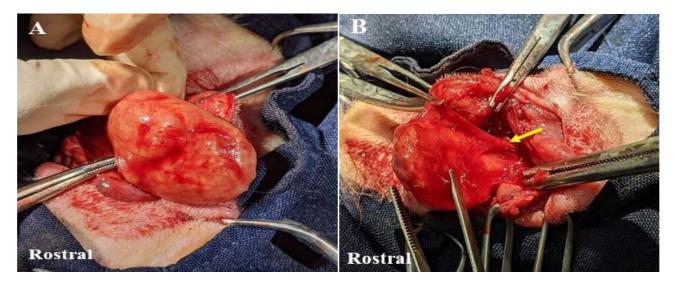


Fig. 5. Close-up view of the (A) lateral surface and the (B) medial surface of the exteriorized firm salivary gland complex. The irregular shape is consistent with the outline seen in radiograph images (Fig. 2). The contiguous gland/duct complex is visible on the medial side, along with a branch of the Maxillary vein (yellow arrow)

The firm mass was identified as the mandibular and sublingual salivary glands. Salivary leakage from the sublingual salivary gland and the duct complex resulted in an inflammatory response, creating granulation tissue around the involved glands. The granulation tissue extended to the closely associated sublingual gland and the connecting ducts – more visible on the medial side (Fig. 5B). Further rostral dissection uncovered the monostomatic and polystomatic portions of the sublingual gland.

The root duct of the sublingual salivary gland was dissected rostrally until the lingual nerve was visible then the duct was ligated with two separate surgeon's knots, and distally transected. The site of the sialocele was flushed with normal saline solution (NSS) and the surrounding tissues were checked for bleeding before closure (Fig. 6). The cut platysma muscle was sutured together with two interrupted surgeon's knots. The subcutaneous layer was closed with a continuous suture pattern and the wound edges of the skin were apposed using a simple interrupted suture pattern. An absorbable braided polyglactin 910 suture material was used. A Penrose drain was placed to drain the sialocele and to prevent the accumulation of edema fluid in the surgical site. Patient recovery post-surgery was good with no signs of subcutaneous bleeding or swelling.



Fig. 6. The appearance of the surgical site before closure of subcutaneous and skin layers.

Co-amoxiclav (20mg/kg BID) metronidazole (25mg/kg BID) were prescribed as post-operative oral antibiotics. Firocoxib (5mg/kg SID) was prescribed as an NSAID to control inflammation and reduce pain. Dakin's solution was advised to be used as a rinsing solution and mupirocin was prescribed for topical wound management. Wound healing over two weeks post-surgery was good with no dehiscence of sutures. The drain and skin sutures were removed after 14 days (Fig. 7). The patient returned for check-ups monthly, for six months post-recovery, and no recurrence of clinical signs or mass regrowth was observed or reported, with the dog staying clinically healthy.

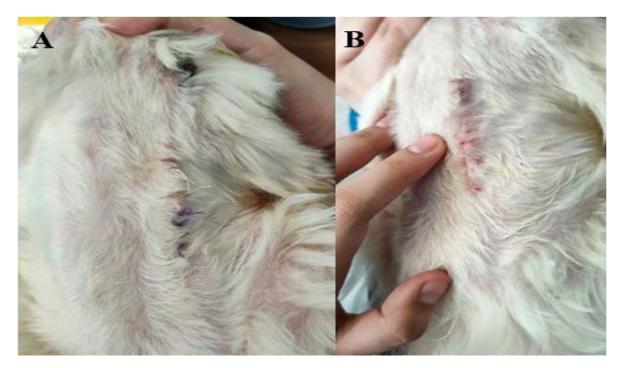


Fig. 7. **Appearance of cervical region 14 days post-operation.** Incision line (A) before and (B) after removal of stitches. The head of the dog is tilted upwards to exhibit the cervical region now free of swelling.

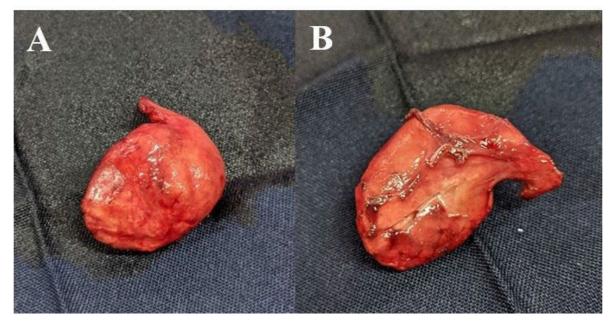


Fig. 8. Excised mass from the cervical region. (A) Lateral surface and (B) medial surface.

The excised mass (Fig. 8) was submitted to a diagnostic laboratory for histopathology processing. Grossly, the mass appeared to be tan-colored, 4.5 x 3.0 x 1.0 cm in size, and irregularly shaped. Cut sections showed a unilocular cyst with cream-white areas of calcification. Microscopically, there was a proliferation of fibrous connective tissue with

small capillaries. The outer layer, adjacent to the epidermis, consisted of areas of calcification that were stained basophilic with a consistent thickness throughout the slide (Fig. 9). The middle layer had interlacing bundles of collagen with fibroblasts scattered throughout. Additionally, this layer had circular bodies of lamellated eosinophilic fibrillar concretions, up to 500 microns in diameter, bound by a thin rim of fibrous connective tissue and fibroblasts. Both the outer layer and middle layer findings indicate sialolith formation. The innermost layer was composed of unremarkable fibrous connective tissue. The pathology report diagnosis is a salivary mucocele or sialocele, focally extensive with granulation tissue, multiple sialoliths, and sialadenitis.

The specific etiologies of a sialocele remain unclear. Previous reports state that traumatic injury is believed to be the inciting cause of a tear in a salivary gland or duct which leads to the formation of a sialocele [2, 15, 16]; however, most documented cases are idiopathic [2, 5].

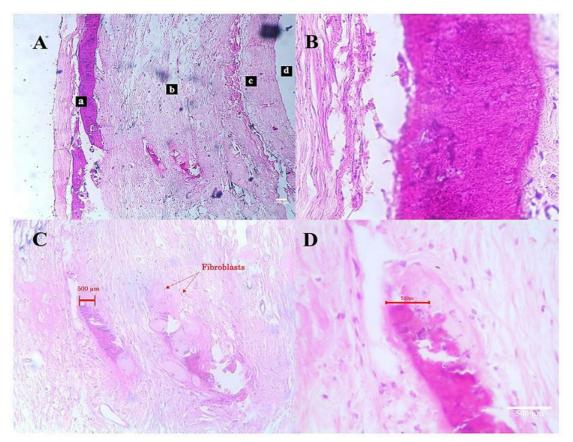


Fig. 9. Photomicrographs of the excised mass. (A) 40x magnification showing the (a) outer layer with marked calcification, (b) middle layer with interlacing bundles of collagen and fibroblasts, (c) inner layer – connective tissue, and (d) inner cavity.; (B) 400x magnification on the outer layer shows eosinophilic granules; (C) and (D) 100x and 400x magnification on the middle layer exhibits the presence of circular bodies of lamellated eosinophilic fibrillar concretions (up to 500 microns in diameter) and is surrounded by fibroblasts (red arrows). 10% neutral buffered formalin was used for tissue preservation. Hematoxylin and eosin stains were used for microscopic examination.

4. Discussion

In dogs, the most common diseases of salivary glands are sialoceles [10]. The site of the sialocele will often determine the presenting clinical signs and indicate which salivary gland or glands it is arising from [9] – as seen in this case of a cervical sialocele with the mandibular and sublingual salivary glands involved.

This case falls under the idiopathic category as the dog was primarily kept indoors and the owner does not recall any event or trauma that may have affected the dog's neck region. Prior accounts by Kazemi *et al.* [8] and Kumar *et al.* [17] indicate that tight pressure and irritation from the neck collar can damage

the mandibular salivary glands and ducts resulting in a leakage and build-up of saliva, forming a sialocele.

Commonly performed treatments for sialoceles include oral medication, fluid drainage, and surgical removal. Oral medication does not treat the root cause and provides only mild relief of discomfort or temporary alleviation of swelling. Simple drainage also does not eliminate the main cause – the defect in the salivary gland and duct complex - and is reported to have a high recurrence rate; therefore, it should be avoided [8, 18, 19]. Drainage can also irritate adjacent tissue and lead to adhesions or scar tissue formation, which can complicate future surgical procedures [8, 18].

sialoceles Since can recur. sialoadenectomy is the definitive and preferred treatment for this condition. Sialoadenectomy is the surgical removal of the entire affected salivary gland and duct complex [20, 21]. In this case, the sialocele was located ventro-caudally to the caudal ramus of the left mandible. This cervical sialocele possessed firm borders due to the formation of extensive granulation tissue in reaction to salivary leakage. The lateral approach technique (LAT) [7] was employed to create ease of access and visualization of the ventrolateral cranial neck region during surgery. It is recommended to remove both mandibular and sublingual salivary glands when one is suspected to be the root of a sialocele [7]. To ensure full removal of the defect, ligate and excise the mandibular and sublingual glands and connected ducts, transecting always caudal of the lingual nerve [4, 22]. King and Waldron [22] state that any defects of the duct occurring rostral to the lingual nerve require no surgical dissection. Complete excision of the mandibular and sublingual salivary glands, formed pseudocysts, and redundant tissues is the most definitive treatment for sialoceles [3, 8].

Possible complications post-sialoadenectomy include general postoperative problems such as seromas, infections, and sialocele recurrences [10]. A study by Cinti *et al.* [7] on the complications between ventral and lateral approach sialoadenectomy reported a 24% postoperative complication rate, with most being seromas and surgical site swellings. Contributing factors are surgical dissection

technique, dead space creation, and the highly mobile location of the incision. To prevent these complications, a Penrose drain was placed in the cervical surgical site before closure. The drain minimizes the formation of seromas and abscesses [9]. Careful identification of the affected glands and ducts with prudent tissue dissection and aseptic technique during surgery should minimize complications, prevent infections, and avoid recurrence [2].

5. Conclusion

The most appropriate treatment for a sialocele is sialoadenectomy - surgical removal of the defect in the affected salivary glands and salivary ducts. Complete sialoadenectomy and removal of related inflammatory tissues, performed with proper technique and aseptic technique, resolves the problem and prevents recurrence. A cervical sialocele, as shown in this case, can present as an abnormal swelling in the neck region that is fluctuant and painless. An untreated sialocele can cause prolonged tissue irritation resulting in the formation of thickened granulation tissue and a firm cervical mass. Diagnosis of a cervical sialocele can be done through physical examination of the affected region, needle aspiration, diagnostic imaging, and cytology. Once diagnosis is made, prompt treatment can prevent chronic or recurring cases of sialoceles. With sialoadenectomy, the prognosis is excellent with good long-term resolution of clinical symptoms.

Author Contributions

Conceptualization, M.B.T.C; Methodology and Investigation, M.B.T.C. and M.I.M.M; Resources, M.B.T.C; Writing – Original Draft, M.B.T.C. and M.I.M.M; Writing – Review & Editing, M.B.T.C.

Ethics Approval and Consent to Participate

This report describes a case that was handled as one of the UP veterinary teaching hospital's routine clinical caseloads; therefore, the institutional animal care and use committee (IACUC) or approval from an ethical governing body was not required. Owner consent was obtained for patient care in all aspects.

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The authors received no specific funding for this work.

Conflict of Interest

The authors declare no conflicts of interest.

Abbreviations

CBC, complete blood count; ALT, alanine aminotransferase; ALP, Alkaline phosphatase; AST, aspartate transaminase; NSS, normal saline solution; NSAID, non-steroidal anti-inflammatory drugs; LAT, lateral approach sialoadenectomy; IM, intra-muscular; SC, subcutaneous; IV, intravenous.

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