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12 **SUMMARY** 

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### 14 **Objectives**

To describe the presentation, diagnosis, cause, complications and outcome in 14 dogs thatpresented with a parotid sialocele and that were treated with complete parotidectomy.

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# 18 Material and Methods

19 Multi-institutional retrospective study.

20 Results:

21 All patients presented with a non-painful, fluctuant, soft tissue mass over the lateral aspect of 22 the face in the region of the parotid salivary gland. A diagnosis was made by means of 23 sialoradiography(3/14), CT(3/14), ultrasound(11/14) and MRI(2/14). The cause of the 24 sialocele could be determined in 8 out of 14 patients and included: foreign body(2/14), 25 sialolithiasis(1/14), neoplasia(3/14), salivary gland lipomatosis(1/14) and trauma(1/14). One 26 anaesthetic complication (regurgitation) and 7 postoperative surgical complications were 27 recorded (self-limiting formation(2/14), haemorrhage(1/14), seroma wound 28 dehiscence (1/14), abscessation 7 months postoperatively due to a retained part of a penrose 29 drain(1/14) and facial nerve paralysis(2/14). None of the patients had recurrence of the 30 sialocele with a median follow-up time of 14 months.

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# 32 Clinical significance:

Parotidectomy has historically been considered a technically challenging procedure but we report that in the hands of an experienced surgeon it can have a good success rate with longterm resolution of the clinical symptoms. Despite this, intra- and postoperative complications can occur and owners should be warned of this prior to surgery.

#### 37 **INTRODUCTION**

A sialocele is a collection of saliva within the subcutaneous tissue caused by leakage from the salivary gland or more commonly its associated duct. These saliva-filled cavities are lined by inflammatory tissue rather than epithelial cells and therefore are not true cysts (Torad and Hassan 2013). The majority of sialoceles are idiopathic and other causes such as trauma, sialoliths, foreign bodies and neoplasia have been described (Waldron and Smith 1991). One patient developed a cervical sialocele as a result of dirofilariosis (Henry 1992).

The location of the sialocele typically determines the presenting complaint and gives a good indication of the offending salivary gland with the submandibular and sublingual glands being most frequently affected followed by the zygomatic salivary gland (Dunning 1985). Reports of parotid sialoceles are uncommon (Goldsworthy and others 2013, Guthrie and Hardie 2014, Harvey 1977, 1981, Jeffreys and others 1996, Ladlow and Gregory 2003, Termote 2003, Trumpatori and others 2007).

Surgery is the treatment of choice for sialoceles as treatment with drainage alone typically results in recurrence in 40% of affected animals. (Waldron and Smith 1991). Several surgical treatments have been proposed for parotid sialoceles including parotidectomy (Guthrie and Hardie 2014, Trumpatori and others 2007), ligation of the duct proximal to the defect (Goldsworthy and others 2013, Harvey 1977), anatomical reconstruction of the duct defect (Harvey 1977, Jeffreys and others 1996, Termote 2003) and marsupialisation (Ladlow and Gregory 2003).

According to Ritter and Stanley (2012), parotidectomy is a technically challenging procedure because of the intimate association of the parotid capsule with its surrounding structures. Dunning (1985) reports a high risk of postoperative complications including iatrogenic facial nerve damage but there is currently little information on postoperative complications in the literature. To the authors' knowledge, only two individual case reports of parotid sialoceles treated with parotidectomy have been published and only one reports on postoperative facial
nerve function (Guthrie and Hardie 2014, Trumpatori and others 2007). Therefore, information
on the risk of parotidectomy in the veterinary literature is currently lacking.

65 The purpose of this study was to describe the presentation, diagnosis, cause, complications and 66 outcome in 14 patients that presented with a parotid sialocele and that were treated with 67 complete parotidectomy.

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### 69 MATERIAL AND METHODS

70 Medical records of 6 referral practices were retrospectively reviewed for the condition of parotid sialocele over the period from 2007 to 2014. Patients were included if the condition was 71 72 treated using a parotidectomy as described by Ritter and Stanley (2012) with a minimum of 6 73 months follow-up. All procedures were performed or supervised by a Diplomate of the 74 European College of Veterinary Surgeons. The following data were obtained: patient 75 identification, breed, age, weight, sex, presenting clinical symptoms, duration of symptoms prior 76 to diagnosis, diagnostic imaging, aetiology, results of cytology if performed pre-operatively, 77 culture and sensitivity results, intra- and postoperative complications, histopathology of 78 resected gland, recurrence and outcome. Descriptive statistical analysis was performed using a 79 statistical software package (Excel for Mac, Microsoft Corporation). Major complications were 80 defined as those requiring further treatment (surgical or non-surgical) whereas minor 81 complications where self-limiting and did not require further treatment. Patients were classed 82 as having postoperative facial nerve paralysis if there was no facial nerve function present 6 83 months postoperatively.

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#### 85 <u>RESULTS</u>

86 Signalment

Fourteen cases met the inclusion criteria (Table 1). The mean age of all patients was 7.8 years
(SD 3.5 years). There were 7 female and 7 male dogs. Neutering status was known for all dogs
and 11 out of 14 patients were neutered. There were 3 boxers, 1 terrier cross, 1 Rottweiler, 1
basset hound, 2 crossbreeds, 1 bearded collie, 1 British bulldog, 1 Jack Russell terrier, 1 saluki
and 2 springer spaniels.

92

# 93 Clinical symptoms

The duration of the clinical signs before referral ranged from 7 to 760 days with a mean of 117 and a median of 30 days. All patients presented with a non-painful, fluctuant, soft tissue mass over the lateral aspect of the face across from the masseter muscle to the parotid region. Other clinical signs were: pain on opening of the mouth and inability to open the mouth fully (n=1), presence of a firm mass in the parotid region associated with the sialocele (n=3) and intra-oral swelling and sinus at the level of the 4<sup>th</sup> pre-molar (n=1). All patients had normal facial nerve function on presentation.

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## 102 Diagnostic Imaging

103 The most commonly used diagnostic modality in this case series was ultrasound (11/14 104 patients) followed by sialography (3/14), computed tomography (CT) (3/14) and magnetic 105 resonance imaging (MRI) (2/14). Three patients had standard 3-view thoracic radiographs to 106 rule out pulmonary metastasis and in one patient sialoradiography was attempted 107 unsuccessfully prior to CT (Table 1). On ultrasound, all sialoceles appeared as round to tubular 108 echogenic structures with various amounts of central anechoic content and a hyperechoic wall. 109 A foreign body was identified in one patient as an echogenic area with posterior acoustic 110 shadowing and in another patient a suspected adenocarcinoma of the parotid gland manifested 111 itself as a calcified structure at the base of the ear canal. CT was performed in 2 patients and this revealed a fluid filled cavity associated with the parotid salivary gland. In one patient small, mineralised bodies could be seen in the dependent part of the cavity compatible with sialoliths (Fig 1). MRI was used in 2 patients which identified a well-defined collection of fluid ventral to the ear in the region of the parotid gland. This fluid extended rostrally in both patients along the cheek following the path of the parotid duct (Fig 2). On T1-weighted images post contrast injection there was enhancement of the wall of the parotid lesion compatible with a sialocele (Fig 3) and in one patient an enhancing mass of the parotid gland was present.

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120 Aetiology

The cause of the sialocele could be determined in 8 out of 14 patients (Table 1) and was: foreign body (2/14), sialolithiasis (1/14), neoplasia (3/14), salivary gland lipomatosis (1/14) and trauma (1/14). The trauma patient had been attacked 7 days prior to referral by a badger and presented with a soft tissue swelling over the lateral aspect of the face and multiple puncture wounds.

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### 127 Cytology

Twelve patients had a fine needle aspiration (FNA) of the mass performed prior to surgery. In all patients, the aspirated fluid was consistent with saliva confirmed by a positive reaction with a mucus-specific stain and in 2 cases there was evidence of a suppurative inflammation with rods and cocci on cytological examination. Culture and sensitivity testing was performed in the patient that was attacked by a badger and this revealed profuse growth of Klebsiella Oxytoca. FNA of the three patients with parotic gland adenocarcinomas failed to identify neoplastic cells in one patient. Instead, adipose tissue with secondary neutrophilic inflammation was found.

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136 Surgery and Complications

All patients underwent parotidectomy and sialocele drainage using a standard technique as
described by Ritter and Stanley (2012) (Fig 4). One anaesthetic and 7 postoperative surgical
complications were recorded in 5 patients (Table 1). All patients survived at least 6 months after
surgery.

141 The intra-operative, anaesthesia complication consisted of a dog that regurgitated during the 142 anaesthesia and developed aspiration pneumonia, which was diagnosed by clinical examination 143 and radiography. This was treated with a combination of potentiated amoxycillin (Synulox; 144 Pfizer) and metronidazole (Metronidazole; Crescent Pharma Ltd.); the patient improved after 145 several days of hospitalisation and made a full recovery.

There were 7 postoperative complications in 5 patients of which 2 were classed as minor and 5 as major complications. The 2 minor complications were self-limiting seromas. The major complications consisted of postoperative haemorrhage necessitating revision surgery but not requiring a blood transfusion (1/14), wound dehiscence (1/14) and abscessation 7 months postoperatively (1/14). Two patients (14%) had postoperative, permanent facial nerve paralysis and none of the patients had recurrence of the clinical symptoms with a mean followup of 14 months.

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## 157 Histology

Histology of the resected parotid gland was performed in 13 out of 14 patients. Eight of the 13 submitted parotid glands had varying degrees of inflammatory changes and 3 had undergone neoplastic transformation in the form of adenocarcinomas; there was one salivary gland lipomatosis and one cystic hyperplasia with fibroplasia and histiocytic inflammation. 162

### 163 **DISCUSSION**

164 The diagnosis of a sialocele is based on the location and fine needle aspiration of the mass, which 165 typically reveals viscous, golden brown or blood-tinged fluid. Aspirated fluid has a low cell count 166 and reacts positively with a mucus-specific stain such as the periodic acid-Schiff, confirming the 167 diagnosis (Smith 2000). A thorough physical examination usually further denotes the origin of 168 the sialocele and all patients in this case series had a typical swelling on the lateral aspect of the 169 face. In all of our cases, further imaging was performed to investigate the cause of the sialocele. 170 Historically, sialography has been the diagnostic modality of choice to determine the extent and 171 cause of the disease process but this requires general anaesthesia and it can be difficult to locate 172 the duct opening (Smith 2000). If sialography is unsuccessful due to obstruction of the duct or 173 difficulty in cannulating the papilla, advanced imaging can be considered. Ultrasonography and 174 CT have been used as diagnostic modalities and both have been found to be useful in identifying 175 the offending salivary gland and cause of the sialocele (Kneissl and others 2011, Torad and 176 Hassan 2013, Trumpatori and others 2007). In this case series, ultrasound was used more 177 frequently than CT and it was found to be useful in identifying 2 foreign bodies and one 178 suspected neoplasm of the parotid gland. Computed tomography was able to identify a sialolith 179 in one case but no cause could be identified in the 2 other patients. In humans, imaging 180 modalities frequently used to diagnose sialolithiasis include sialography, sialoendoscopy, 181 ultrasonography and more recently CT and MRI (Avrahami and others 1996, Jager and others 182 2000). Sialoendoscopy has become a commonly performed procedure in people but it is not 183 clear if it can be performed in dogs (Nahlieli and Baruchin 1999, Trumpatori and others 2007). 184 To the author's knowledge, this is the first time that MRI has been used in the diagnosis of a 185 parotid sialocele. MRI was useful to identify underlying inflammation of the parotid gland and 186 the extent of the disease process.

187 Surgery is the treatment of choice for sialoceles and in most cases excision of the offending gland 188 is curative and carries a good prognosis with a low risk of postoperative complications (Waldron 189 and Smith 1991). In the case of a parotid sialocele, the surgical removal of the parotid gland is 190 more challenging because of the intimate association of the parotid capsule with its surrounding 191 structures (Ritter and Stanley 2012). Therefore other surgical treatments have been used to 192 address the problem. Anatomical reconstruction of the defect and duct has been described but 193 this relies on the ability to identify and suture the defect (Harvey 1977, Jeffreys and others 1996, 194 Termote 2003). There are also concerns that that duct would not remain patent after 195 reconstruction (Trumpatori and others 2007). Alternatively, the duct can be ligated proximal to 196 the defect as described by Goldsworthy and others (2013) causing atrophy of the gland. Ligation 197 of the proximal parotid duct results in progressive glandular atrophy(Harrison and Garrett 198 1976). This technique produces consistent atrophy of the parotid gland in comparison to the 199 mandibular and sublingual glands which may be related to the predominance of serous cells in 200 the acini of the parotid gland (DeYoung and others 1978). Intra-oral marsupialisation of the 201 distended duct is a simple technique but can only be performed in patients where the sialocele 202 extends to the buccal mucosa. None of the techniques described above would be suitable to deal 203 with a parotid sialocele secondary to a neoplastic process.

204 Dunning (1985) describes parotidectomy for the treatment of parotid sialoceles as a challenging 205 procedure carrying a high risk of complications and permanent facial nerve paralysis but there 206 is currently little evidence in the literature to support this opinion. We report a moderate 207 complication rate in our series with 5 out of 14 patients (35%) developing a major complication 208 requiring further treatment. The first patient with a major postoperative complication 209 developed an acute swelling over the incision site 24 hours after surgery. Fine needle aspiration 210 was suggestive of haemorrhage and because the swelling was getting progressively worse and 211 the patient started to bleed from the incision, the decision was made to revise the procedure. 212 During surgery, active bleeding from a branch of the caudal auricular artery was identified. The 213 vessel was ligated and the patient recovered uneventfully without the need for a blood 214 transfusion. The second postoperative complication consisted of wound breakdown requiring 215 surgical debridement and a muscle flap to reconstruct the lip. This patient had been attacked 7 216 days prior to referral by a badger and presented with a soft tissue swelling over the lateral aspect 217 of the face and multiple puncture wounds. Swabs taken for culture and sensitivity testing taken 218 at the time of the parotidectomy found profuse growth of Klebsiella oxytoca and the 219 contamination of the area was likely a contributing factor in the wound breakdown. Another 220 patient developed an abscess 7 months postoperatively; on exploration of the abscess a small 221 part of a penrose drain was found likely responsible for the abscessation.

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The 2 patients that developed permanent facial nerve paralysis both required revision surgery. However, the facial nerve function was absent after the initial procedure so therefore this can be considered separate from the complication. None of the patients in this study had recurrence of the sialocele with a median follow-up time of 14 months and thus parotidectomy compares favourably with other surgical techniques in that respect. To our knowledge this is the first time that the complication rate for this procedure has been quantified in a larger group of patients.

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This study had some major limitations. First of all, the retrospective nature of this study made it difficult to get all the relevant information for all the patients. This is especially true because this is a multi-institutional study and not all institutions are equally proficient in retrieving patient information. Secondly, the small number of patients reduces the statistical power of this study and because of the disparity of aetiologies in the current case series, any conclusion on outcome could be biased. The main reason for this is that parotid sialoceles are uncommon and several treatment options are available to deal with it. Only 14 cases were identified over a period of 7
years across 6 busy referral hospitals.

Furthermore, not all procedures were carried out by the same surgeon and this difference insurgical experience could have given rise to different outcomes.

Despite all of these shortcomings, the authors feel that this study brings useful information to the scientific community and can help surgeons in their decision-making and client communication when they come across this unusual condition.

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## 245 <u>CONCLUSION</u>

Although historically complete parotidectomy for the treatment of a parotid sialocele has been considered a technically challenging procedure with a high complication rate, this study has found that in the hands of an experienced surgeon the procedure has a good success rate with good long-term resolution of the clinical symptoms.

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# 255 **CONFLICTS OF INTEREST**

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257 Figure legend:

Figure 1. Transverse CT image of the head taken after intra-venous contrast injection. A large,

well-delineated hypo-attenuating fluid-filled structure can be seen in the ventral aspect of the

- 260 head consistent with a parotid sialocele. Several small-sized mineralised bodies are seen in the
- 261 dependent part of the lesion consistent with sialoliths
- 262



Figure 2. T2-weighted dorsal image of patient with a parotid sialocele. There is a well-defined collection of fluid ventral to the ear in the region of the parotid gland, which extends rostrally along the cheek following the path of the parotid duct



Figure 3. T1-weighted, post-contrast, transverse MRI image of patient with a parotid sialocele.

A well-defined collection of fluid ventral to the right ear can be seen (\*). There is enhancement of the wall (arrow) of the lesion post-contrast injection consistent with a sialocele. 



- Figure 4. Intra-operative photograph demonstrating the intimate association of the parotid capsule with its surrounding structures including the maxillary and temporal arteries



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