CASE REPORT

Diagnosis and treatment of a caecal mucocoele in a dog

M. McKenna1*, A. Suárez-Bonnet2†, K. Smith1 and S. Stewart*

1Department of Clinical Science and Services, Royal Veterinary College, North Mymms, Hertfordshire AL9 7TA, UK
2Department of Pathobiology and Population Sciences, Royal Veterinary College, North Mymms, Hertfordshire AL9 7TA, UK

Accepted: 9 August 2019

Introduction

Appendiceal mucocoeles are a well-described entity in humans. The reported incidence in humans is 0.2 to 0.7% of all excised appendices (Marudanayagam et al. 2006), with an increased incidence reported in females compared to males (4:1) and in those over 50 years of age (Aho et al. 1973). Appendiceal mucocoele is a collection term for all cases where an accumulation of mucus due to appendiceal obstruction is present (Stocchi et al. 2003). Appendiceal mucocoeles in humans are classified into one of three types, according to the histologic characteristics of the epithelium (Stocchi et al. 2003):

1. simple mucocoele: defined as appendiceal dilation and accumulation of mucus secondary to obstruction of appendiceal outflow.
2. Benign mucocoele (cystadenoma): defined as a diluted, mucus-filled appendix lined by adenomatous mucosa.
3. Malignant mucocoele (cystadenocarcinoma): defined as the presence of an adenocarcinoma with a dilated, mucus-filled appendix.

Appendiceal mucocoeles may present as incidental surgical findings or may cause a variety of clinical signs including abdominal pain, a palpable abdominal mass and weight loss (Stocchi et al. 2003). In cases of malignant mucocoeles, some neoplastic cells may penetrate the appendiceal wall and seed the peritoneum, leading to accumulation of adhesive, semi-solid mucin in the abdominal cavity in which neoplastic cells can be found; this condition is termed pseudomyxoma peritonei. Retropertitoneal and pleural seeding has also been sporadically reported (Stevens et al. 1997, Takahashi et al. 1998, Peek & Beets 1999). Both benign and malignant mucocoeles may lead to appendiceal perforation (Gibbs 1973).

The following databases (PubMed, Ovid, ScienceDirect) and journals (Veterinary Surgery, Journal of Veterinary Internal Medicine, Journal of Small Animal Practice, Journal of the American Veterinary Medical Association) were searched using the keywords “cecal mucocoele” OR “caecal mucocoele” AND “dog” on August 3, 2020. One report (Phillips & Aronson 2012) of a caecal mucocoele was found. In that case, the caecal mucocoele was identified as an apparently incidental finding during diagnostic investigations for complications of a previous renal transplantation. Typhlectomy was performed and the dog was ultimately euthanised due to the development of post-operative septic peritonitis. One case of pseudomyxoma peritonei (Bertazzolo et al. 2003) has also been reported.

Case history

An 11-year-old male intact Staffordshire Bull terrier was referred for diabetic ketoacidosis. Abdominal ultrasonographic examination revealed a 5 cm × 2 cm intraluminal caecal mass-like structure. Exploratory laparotomy and typhlectomy were subsequently performed. Histopathology of the caecal mass-like structure was consistent with a caecal mucocoele, defined as a cystic dilation of the caecal lumen with stasis of mucus. This lesion has been previously described in humans, where it is termed an appendiceal mucocoele. The patient was euthanased 58 days post-operatively due to unrelated diabetic complications.

An 11-year-old male intact Staffordshire Bull terrier was referred for diabetic ketoacidosis. Abdominal ultrasonographic examination revealed a 5 cm × 2 cm intraluminal caecal mass-like structure. Exploratory laparotomy and typhlectomy were subsequently performed. Histopathology of the caecal mass-like structure was consistent with a caecal mucocoele, defined as a cystic dilation of the caecal lumen with stasis of mucus. This lesion has been previously described in humans, where it is termed an appendiceal mucocoele. The patient was euthanased 58 days post-operatively due to unrelated diabetic complications.

An 11-year-old male intact Staffordshire Bull terrier was referred for diabetic ketoacidosis. Abdominal ultrasonographic examination revealed a 5 cm × 2 cm intraluminal caecal mass-like structure. Exploratory laparotomy and typhlectomy were subsequently performed. Histopathology of the caecal mass-like structure was consistent with a caecal mucocoele, defined as a cystic dilation of the caecal lumen with stasis of mucus. This lesion has been previously described in humans, where it is termed an appendiceal mucocoele. The patient was euthanased 58 days post-operatively due to unrelated diabetic complications.

An 11-year-old male intact Staffordshire Bull terrier was referred for diabetic ketoacidosis. Abdominal ultrasonographic examination revealed a 5 cm × 2 cm intraluminal caecal mass-like structure. Exploratory laparotomy and typhlectomy were subsequently performed. Histopathology of the caecal mass-like structure was consistent with a caecal mucocoele, defined as a cystic dilation of the caecal lumen with stasis of mucus. This lesion has been previously described in humans, where it is termed an appendiceal mucocoele. The patient was euthanased 58 days post-operatively due to unrelated diabetic complications.
were performed to screen for inciting causes of diabetic ketoacidosis. On abdominal ultrasound, a focal tubular intraluminal mass-like structure measuring 5 cm × 2 cm was found in the region of the ileocaecocolic junction (Fig 1A, B), deemed to be caecal in origin. The central portion of the mass-like structure appeared irregular, displaying mixed echogenicity and occasional mineralised foci. No evidence of vascularisation was detected within the mass-like structure on colour Doppler examination. The remainder of the caecum was considered to be normal in appearance, with no loss of wall layering. No obvious continuity with the caecal wall was noted. No concurrent focal steatitis or right colic lymphadenopathy was noted. The rest of the abdominal ultrasonographic examination was within normal limits, except for mildly reduced corticomedullary definition in both kidneys.

Due to concerns that the caecal mass-like structure could be a gastrointestinal stromal tumour (GIST), which in humans has been documented to cause hypoglycaemia due to production of insulin-like growth factors (Davda & Seddon 2007), or a possible cause of insulin resistance contributing to diabetic ketoacidosis, exploratory laparotomy was performed. Grossly, the caecal lesion appeared as an intra-luminal 6.5 × 3.5 × 3.0 cm mottled, beige, discrete, mass-like lesion (Fig 2A). There was marked distension of the caecal lumen and extensive ulceration of the mucosa without evidence of rupture. Total typhlectomy was performed by placement and activation of a transverse stapling device at the base of the caecum. Diffuse heterogeneous nodules were visualised throughout the liver. A suture guillotine method was used to biopsy the margin of an affected liver lobe. The remainder of the exploratory laparotomy was unremarkable; the pancreas was grossly normal and no lymphadenomegaly was noted. Recovery from anaesthesia was smooth and uneventful.

On histopathology, residual intact caecal mucosa was simple to pseudo-stratified columnar rather than the expected cryptal architecture and was variably atrophic (Fig 2B, C and D). The caecal lumen was plugged by acidophilic proteinaceous and cholesterol-rich debris interspersed with mucus, intact and lysed erythrocytes and coarsely granular haemosiderophages. The submucosa and tunica muscularis were variably thinned with areas of fibroplasia, osseous metaplasia and granulomatous or lymphocyte-rich inflammation. The serosal layer was intact and congested with scattered recent haemorrhages. Submucosal, external muscular and serosal layers at the surgical margin were within normal limits. As this lesion was unexpected, two additional board-certified pathologists reviewed the case and all confirmed the diagnosis of caecal mucocoele. Liver histopathology was consistent with acute mild portal hepatitis, most likely attributable to metabolic stress.

Three days post-operatively the patient became inappetent, lethargic and began vomiting. Repeat abdominal ultrasound revealed the pancreas now appeared mildly enlarged, generally hypoechoic and heterogenous, suggesting the development of post-operative pancreatitis. Following intensive supportive care with intravenous fluid therapy and intravenous maropitant (1 mg/kg every 24 hours), ondansetron (0.5 mg/kg every 12 hours) and omeprazole (1 mg/kg every 12 hours), the patient's clinical signs resolved. He was transitioned back on to subcutaneous insulin and discharged 8 days after surgery. The patient re-presented a subsequent eight times over the following weeks due to repeated bouts of diabetic ketoacidosis and pancreatitis. He was subsequently also diagnosed with exocrine pancreatic insufficiency, based on a trypsin-like immunoreactivity result of 3 g/L (reference interval 6-35 g/L). The patient was euthanased 58 days post-operatively due to quality of life concerns associated with poorly-controlled diabetes mellitus and chronic pancreatitis. Post-mortem examination was declined.

DISCUSSION

In human medicine some patients with caecal mucocoeles present with abdominal pain, a palpable abdominal mass, vomit-
Caecal mucocoele in a dog

However, approximately 50% of human cases are diagnosed as incidental findings (Merran 1997, Pickhardt et al. 2002). Although there was initially concern that the caecal mass-like lesion in our patient could have been a contributing factor to the patient’s diabetic ketoacidosis and/or historical suspected hypoglycaemic episodes, given the subsequent diagnosis of caecal mucocoele, it is ultimately deemed to have been an incidental finding in our canine patient. The gross and histopathological findings in this case were most consistent with a simple mucocoele. No adenomatous change or cellular atypia were observed, which consequently rules out a benign mucocoele (cystadenoma) and malignant mucocoele (cystadenocarcinoma).

In human medicine, pre-operative diagnosis of malignant appendiceal mucocoeles is considered important for the selection of surgical methods to prevent peritoneal dissemination, peri-operative complications and the possible need for repeat surgery (Dhage-Ivatury & Sugarbaker 2006, Sugarbaker 2009a). In 2003 Stocchi et al. described clinical, diagnostic and surgical variables associated with malignancy of appendiceal mucocoeles in humans. They found that there was significant association between the presence of symptoms (particularly abdominal pain and weight loss) with malignancy. Other variables associated with malignancy included the presence of a palpable abdominal mass, and the presence of pseudomyxoma peritonei or mucocoele extravasation. Mucocoele size was not associated with malignancy, however, cystadenomas were typically significantly larger than simple mucocoeles. Documentation of additional cases of canine caecal mucocoeles is needed to determine whether such variables are significant in dogs. Abdominal ultrasound, CT, and colonoscopy are the primary means of diagnosis of appendiceal mucocoeles described in humans. The ultrasonographic appearance has been described as a cystic, encapsulated lesion, firmly attached to the caecum, with liquid content and an internal variable echogenicity related to mucus density (Zissin et al. 1999). CT is regarded as the gold-standard for diagnosis of appendiceal mucocoeles in humans, with characteristic CT findings including an appendiceal lumen >13 mm diameter, with cystic dilatation and calcification of the appendiceal walls (Pickhardt et al. 2002, Francica et al. 2006, Ruiz-Tovar et al. 2007, Bennett et al. 2009, Lozano et al. 2010). Colonoscopy may reveal the presence of yellow discharge at the appendiceal orifice, and in some cases a pathognomonic “sign of the volcano,” i.e. a mass with a central crater, from which mucus is discharged (Hamilton & Stormont 1989).

In our canine patient, a caecal mass-like structure was visualised on abdominal ultrasound. Our top differential diagnosis for the caecal mass-like structure was a GIST, which in humans have been documented to cause hypoglycaemia due to production of insulin-like growth factors (Davda & Seddon 2007) and could have explained the suspected hypoglycemic episodes noted before presentation. Most caecal masses in dogs are malign-
nant, with commonly reported diagnoses including GIST, leiomyosarcoma and leiomyoma (Maas et al. 2007). In humans, ultrasonographic features of GISTs include a large extramural tumour arising from the muscularis layer, that rarely affects the mucosal layer, and often contain regions of caviation (Wronski et al. 2009), however, species differences in anatomy limit extrapolation of human imaging findings to canine patients. In veterinary medicine, ultrasonographic characteristics of smooth muscle tumours include eccentric masses with caviations, but these were described before our understanding of GISTs (Myers & Penninick 1994) as a distinct entity. A more recent publication did not show a statistically significant difference between GISTs and non-GISTs using the ultrasonographic features of echogenicity, echotexture, size, bowel wall distribution/morphology or caviation (Hobbs et al. 2015). A caecal mucocoele was not considered a differential diagnosis for the caecal mass-like lesion seen on ultrasound in our patient, given the paucity of reported cases in dogs. This case shows that mucocoele should be considered as an additional differential diagnosis for a canine caecal mass-like structure. The ultrasonographic appearance of the caecal mucocoele in this patient shares some common features with the described ultrasonographic appearance in humans, including an internal mixed echogenicity (Zissin et al. 1999) and lack of vascularisation on Doppler examination (Paladino et al. 2014, Panagopoulos et al. 2017), features which may help distinguish mucocoeles from neoplastic causes of caecal mass-like lesions. Documentation of additional cases of caecal mucocoeles is needed to further characterise the imaging findings and optimal means of diagnosis in dogs.

Exploratory laparotomy and typhlectomy were performed in our patient due to concerns that the caecal mass-like structure could be a GIST. In humans, exploratory laparotomy is preferred to laparoscopy to decrease the risk of mucocoele rupture and pseudomyxoma peritonei, and to facilitate full inspection of the abdominal cavity (Dhage-Ivatury & Sugarbaker 2006, Karakaya et al. 2008, Khan et al. 2010, Sugarbaker 2009a). Though abdominal exploration is vital, as appendiceal mucocoeles have been associated with other tumours in people, particularly colonic adenocarcinomas and ovarian tumours (Kahn & Friedman 1979). In humans appendectomy is indicated for simple mucocoeles, mucosal hyperplasia and for most cystoadenomas. Typhlectomy is indicated for cystoadenomas where appendectomy alone is not surgically feasible while right hemi-colectomy is recommended for cystoadenocarcinomas (Kahn & Friedman 1979, González Moreno et al. 1998). In our patient, total typhlectomy was performed. Due to the paucity of other canine cases, and anatomical differences between species, the optimal surgical management of caecal mucocoeles in dogs is unknown. However, typhlectomy is advised given the reports of spontaneous appendicetal rupture in humans (Gibbs 1973).

Postoperatively, human patients with benign mucocoeles have an excellent prognosis with 5-year survival rates of 91–100%. However 5-year survival rates in cases of malignant mucocoeles are significantly lower (25%) due to complications of pseudomyxoma peritonei (Aho et al. 1973). As our patient’s mucocoele was a simple mucocoele, we anticipate his prognosis would have been excellent in the absence of concurrent disease, however documentation of further canine cases is needed to more accurately determine prognosis is dogs with this condition.

In conclusion, caecal mucocoele should be considered as a differential diagnosis for a caecal mass in canine patients. The detection of a non-vascularised structure with internal mixed echogenicity within the caecum on ultrasound examination should raise suspicion for a possible mucocoele. Based on human literature, exploratory laparotomy and typhlectomy are indicated for suspected caecal mucocoeles, although description of further cases is needed to determine the optimal means of diagnosis and management, and to determine the prognosis of these cases.

Acknowledgements

The authors would like to thank Dr. G.A. Ramirez, DVM, PhD, Dipl ECVP, for reviewing the case and for his constructive comments, and Dr. Helen Dirrig, BVetMed, MvetMed, Dipl ACVR, Dipl ECVDI, MRCVS, for providing the ultrasound images.

Conflict of Interest

None of the authors of this article has a financial or personal relationship with other people or organisations that could appropriately influence or bias the content of the paper.

References

Aho, A., Heinonen, R. & Laureen, P. (1973) Benign and malignant mucocoele of the appendix. Acta Chirurgica Scandinavica 139, 392-400

Benett, G. L., Tanpitukpongr, T. P., Macari, M., et al. (2009) CT diagnosis of mucocoele of the appendix in patients with acute appendicitis. American Journal of Roentgenology 192, 103-110

Bertazzolo, W., Roccabianca, P., Crippa, L., et al. (2003) Clinicopathological evidence of pseudomyxoma peritonei in a dog with intestinal mucinous adenocarcinoma. Journal of the American Hospital Association 39, 72-75

Davda, R. & Seddon, B. M. (2007) Mechanisms and management of non-islet cell hypoglycaemia in gastrointestinal stromal tumour: case report and a review of published studies. Clinical Oncology 19, 265-268

Dhage-Ivatury, S. & Sugarbaker, P. H. (2006) Update on the surgical approach to mucocoele of the appendix. Journal of the American College of Surgeons 202, 680-684

Francisco, G., Lapicicrella, G., Giardibello, C., et al. (2006) Giant mucocoele of the appendix: clinical and imaging finding in 3 cases. Journal of Ultrasound in Medicine 25, 643-648

Garcia Lozano, A., Vazquez Tarrago, A., Castro Garcia, C., et al. (2010) Mucocoele of the appendix: presentation of 31 cases. Cirugia Espanola 87, 108-112

Gibbs, N. M. (1973) Mucinous cystadenoma and cystadenocarcinoma of the vermiform appendix with particular reference to mucocoele and pseudomyxoma peritonei. Journal of Clinical Pathology 26, 413-421

González Moreno, S., Shinmooker, B. M. & Sugarbaker, P. H. (1998) Appendiceal mucocoele. Contraindication to laparoscopic appendectomy. Surgical Endoscopy 12, 1177-1179

Hamill, D. L. & Stormont, J. M. (1989) The volcano sign of appendiceal mucocoele. Gastrointestinal Endoscopy 35, 453-456

Hobbs, J., Sutherland-Smith, J., Penninck, D., et al. (2015) Ultrasonographic appearance of canine gastrointestinal stromal tumors compared to other gastrointestinal spindle cell tumours. Veterinary Radiology & Ultrasound 56, 432-438

Kahn, M. & Friedman, I. H. (1979) Mucocoele of the appendix: diagnosis and surgical management. Diseases of the Colon and Rectum 22, 267-269

Karakaya, K., Barut, F., Emre, A. U., et al. (2008) Appendiceal mucocoele: case reports and review of current literature. World Journal of Gastroenterology 14, 2286-2293

Khan, M. R., Ahmed, R. & Saleem, T. (2010) Intricacies in the surgical manage- ment of appendiceal mucinous cystadenoma: a case report and review of litera- ture. Journal of Medical Case Reports 5, 129

Lozano, A. G., Tarrago, A. V., Garcia, C. C., et al. (2010) Mucocoele appendicular: presentacion de 31 casos. Cirugía española 87, 108-112

Maas, C., Ter Haar, G., Van Der Gag, I., et al. (2007) Reclassification of small intestinal and cecal smooth muscle tumors in 72 dogs: clinical, histologic, and immunohistochemical evaluation. Veterinary Surgery 36, 302-313
Marudanayagam, R., Williams, G. T. & Rees, B. I. (2006) Review of the pathological results of 2660 appendectomy specimens. Journal of Gastroenterology 41, 745-749
Merran, S. (1997) Mucus secreting tumor of the appendix (appendiceal mucocoele). La Presse Medicale 26, 533
Myers, N. C. & Penninck, D. G. (1994) Ultrasonographic diagnosis of gastrointestinal smooth muscle tumors in the dog, Veterinary Radiology & Ultrasound 35, 391-397
Paladino, E., Bellantone, M., Conway, F., et al. (2014) Large mucocoele of the appendix at laparoscopy presenting as an adnexal mass in a postmenopausal woman: a case report. Case Reports in Obstetrics and Gynecology 2014, 1-4
Panagopoulos, P., Tsokaki, T., Misiakos, E., et al. (2017) Low-grade appendiceal mucinous neoplasm presenting as an adnexal mass. Case Reports in Obstetrics and Gynecology 2017, 1-3
Peek, D. F. & Beets, G. L. (1999) Pseudomyxoma peritonei in the pleural cavity: report of a case. Diseases of the Colon and Rectum 42, 113-115
Phillips, H. & Aronson, L. R. (2012) Use of end-to-side arterial and venous anastomosis techniques for renal transplantation in two dogs. Journal of the American Veterinary Medical Association 240, 296-303
Pickhardt, P. J., Levy, A. D., Rohrmann, C. A., et al. (2002) Primary neoplasms of the appendix manifesting as acute appendicitis: CT findings with pathologic comparison. Radiology 224, 775-781
Ruiz-Tovar, J., Teruel, D. G., Gastineires, V. M., et al. (2007) Mucocoele of the appendix. World Journal of Surgery 31, 542-548
Smeenk, R. M., van Velthuysen, M. L., Verwaal, V. J., et al. (2008) Appendiceal neoplasms and pseudomyxoma peritonei: a population-based study. European Journal of Surgical Oncology 34, 196-201
Stevens, K. J., Dunn, W. K. & Balfour, T. (1997) Pseudomyxoma extraperitonei: a lethal complication of mucinous adenocarcinoma of the appendix. American Journal of Gastroenterology 92, 1920-1922
Stocchi, L., Wolff, B. G., Larson, M., et al. (2003) Surgical treatment of appendiceal mucocoele. Archives of Surgery 138, 585-590
Sugarbaker, P. H. (2009a) Appendiceal epithelial neoplasms and pseudomyxoma peritonei, a distinct clinical entity with distinct treatments. In: General Surgery. Principles and International Practice. Eds K. J. Bland, M. W. Buchler, A. Csendes, O. Y. Garden, M. G. Saar and J. Wong. Springer, London. pp 885-893
Sugarbaker, P. H. (2009b) Epithelial appendiceal neoplasms. Cancer Journal 15, 225-235
Takahashi, S., Furukawa, T. & Ueda, J. (1998) Case report: mucocoele of the tip of the appendix. Clinical Radiology 53, 149-150
Wronska, M., Cebulski, W., Slodkowski, M., et al. (2009) Gastrointestinal stromal tumors: ultrasonographic spectrum of the disease. Journal of Ultrasound in Medicine 28, 941-948
Zissin, R., Gayer, G., Kots, E., et al. (1999) Imaging of mucocoele of the appendix with emphasis on the CT findings: a report of 10 cases. Clinical Radiology 54, 826-832