Fatal esophageal fibrosarcoma associated to parasitism by spirurid nematode *Spirocerca lupi* in a dog: a case report

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Abstract A 7-years-old intact female mixed breed dog was referred to the private veterinary clinic because of history of intense food regurgitation. The plain X-ray examination and the contrast-enhanced X-ray using barium sulfate revealed the presence of a radio-opaque mass in the final third of the esophagus. Stool samples were collected and the coproparasitological test was done, which showed thick-shelled eggs from *Spirocerca lupi*. As the canine died, the necropsy was performed, showing nodular lesions in the esophagus, compatible with the typical lesions of spirocercosis. The histopathological analysis showed the proliferation of spindle-shaped cells, process compatible with fibrosarcoma. This is the first official report of the presence of canine spirocercosis in Guapimirim city, Rio

de Janeiro, Brazil, and can serve as a warning to veterinary practitioners, about this probably endemic region for *S. lupi*.

Keywords Brazil · Dog · Esophageal fibrosarcoma · *Spirocerca lupi*

Introduction

Spirocercosis is a disease caused by the nematode *Spirocerca lupi* (Rudolphi 1819), belonging to Spirurida order and affects especially Canidae, or most rarely Felidae family members. When adults, the worms are located in cystic nodules in the esophagus, which communicate with the lumen via fistulas. *S. lupi* eggs contain the L1 larvae, which can be eliminated through gastrointestinal tract into the feces or shed in the vomit (Mense et al. 1992; van der Merwe et al. 2008). Eggs can be ingested by intermediate hosts, the coprophagic beetles of Scarabaeidae family and the larvae encyst within the tissues. Beetles are ingested by dogs, the final hosts, or a paratenic hosts such as poultry, lizards, hedgehogs, wild birds, rabbits, and rodents (Bailey et al. 1963; Bailey 1972; Fox et al. 1988). The infecting larvae L3, when eaten by dogs, excyst in the stomach and penetrate the gastric mucosa, after migrating through the arteries and later to the esophagus as immature adults (Bailey 1963, 1972; van der Merwe et al. 2008).

Spirocerca lupi is worldwide distributed in warm climate regions and canine cases of spirocercosis have been reported in Brazil (Santos et al. 2004). Spirocercosis has a variety of clinical presentations and parasitized animals can present several complications such as development of aortic lesions, secondary megaesophagus and bacterial infections, spondylitis of the thoracic vertebrae and

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esophageal tumors (Bailey 1963; Dvir et al. 2001; van der Merwe et al. 2008). Esophageal tumors are considered rare in dogs and represent only 0.5 % of canine neoplasms (Ridgeway and Suter 1979).

Case report

A 7-years-old intact female mixed breed dog, fed commercial ration, lived previously as a stray dog and now living off-leash on a farm in the Guapimirim city (22°32'14" S and 42°58'55" W), Rio de Janeiro state, Brazil, was admitted to the private veterinary clinic with history of intense food regurgitation.

The plain X-ray examination revealed the presence of a radio-opaque mass in the final third of the esophagus, with diameters 6.5 cm long and 4.0 cm high (Fig. 1a). In the contrast-enhanced X-ray using barium sulfate, a radio-opaque mass was observed in the final third of the esophagus, with the same dimensions as previously noted and a slight retention of the contrast medium before the mass. In addition, the esophagus was outside of its normal position (Fig. 1b). Given the clinical presentation, radiographic findings, and because there were reported cases of spirocercosis in this region in the late nineties (unofficial data), this was our main clinical suspicion.

Canine stool samples were collected and preserved in vials containing MIF, and later sent for laboratory analysis. The coproparasitological test was done using the routine methods in our laboratory, the Willis and Hoffman techniques and analyzed by optical microscopy bright field. Both tests revealed the presence of thick-shelled eggs, compatibles with *S. lupi* eggs (Fig. 2), thus confirming our suspicions. Even after combined treatment with two doses of ivermectin (600 µg/kg, SC, 2 weeks apart) and

Fig. 2 A thick-shelled *S. lupi* egg containing a L1 larvae (scale bar 35 µm)

prednisolone (0.5 mg/kg, bid for 2 weeks, followed by dose tapering), this dog died and the necropsy was performed.

During the necropsy, the piece constituted by the esophagus was removed, which could be observed nodules on external and internal walls of the esophagus (Fig. 3) and a *S. lupi* worm can be removed from a typical esophageal fistula in the internal nodule. In the serous surface, three nodules up to 2.5 cm were found near the caudal border. One of the nodules showed chestnut brown nematode parasites in the central region, and these nematodes showed typical structures of adult *S. lupi* worms, (Fig. 3, upper), e.g., a pink-reddish cuticle. The other nodules were

Fig. 1 **a** Right lateral thoracic plain X-ray, a mass can be observed in the final third of the esophagus (arrows). **b** Contrast-enhanced X-ray using barium sulfate, a partial retention of the contrast medium can be observed (arrow), before the mass in the final third of the esophagus

Fig. 3 A nodule on internal wall of esophagus (red dashed circle), showing a typical esophageal fistula (black arrow) that communicates a nodule, with the esophageal lumen, and where a *S. lupi* worm (blue arrow) with approximately 4.0 cm of length can be removed (black scale bar 5 cm). The upper figure shows the pink-reddish cuticle, a typical structure this nematode (white scale bar 1.5 cm). (Color figure online)

preserved in vials containing 10 % buffered formalin for later histopathological analysis.

When the histopathological cleavage was performed, the other two nodules showed a firm consistency and had a cut surface, either compact, sometimes fasciculated, with a whitish-yellow tone and whitish spots, and small cystic cavities among them. The histological cuts stained by hematoxylin and eosin showed empty cystic spaces in the center, surrounded by mononuclear and polymorphonuclear inflammatory cells, and thick fibrosis. The lesion compromised the esophageal submucosa, and the scar fibrosis sometimes extended from the mucosal tunic, reaching the submucosa and the muscular tunics. Another observation was the deposition of extracellular matrix between cells and a proliferation of spindle-shaped cells with little pleomorphism and no mitosis, but that formed bundles in swirls, process compatible with fibrosarcoma (Fig. 4).

All the procedures were carried out with permission from the responsible for the animal.

Discussion

We report a fatal case of esophageal fibrosarcoma *S. lupi*-associated in a dog of Guapimirim city, Rio de Janeiro, Brazil.

The diagnosis in life was performed using X-ray examination and coproparasitological tests. According to

Fig. 4 Histological cut stained by Hematoxylin and Eosin showing a typical fibrosarcoma cells. The view shows a deposition of extracellular matrix between cells and a proliferation of spindle-shaped cells with little pleomorphism and no mitosis, but that formed bundles in swirls (scale bar 280 μ m)

Dvir et al. (2001), the radiological findings typical of spirocercosis are a caudal esophageal mass, lesions in the aorta artery, and thoracic spondylitis. The finding of a mass in the chest, located in the caudal mediastinum, obtained in this study by plain radiography, corroborates with this author. The other alterations mentioned were not found. In the work of Gosling and Gracia (1983), the contrast-enhanced X-ray showed the sinuously shaped retention of the contrast medium at the level of the last third of the thoracic esophagus, located upwardly before the cardia or the cardiac silhouette, depending on the severity of the case. In the present study, a partial retention of contrast medium occurred in the portion before the mass, situated in the last third of the esophagus, which also corroborates with these authors.

Modified coproparasitological methods are usually preferred for identification of *S. lupi* eggs such as artificial gastric juice and sugar centrifugal flotation (Cabrera and Bailey 1964; Markovics and Medinski 1996). Other methods such as flotation with sodium nitrate solution or supersaturated 33 % zinc sulfate have been described to improve diagnosis (van der Merwe et al. 2008). Here, the Willis method showed lower efficiency when compared to Hoffman method (one versus five eggs counts, respectively), as previously reported by other authors (Cabrera and Bailey 1964). The Hoffman method proved to be efficient in identifying of *S. lupi* eggs, even though this is a plain (not modified) sedimentation method, and this result is only a single sample.

The post-mortem diagnosis was done through of necropsy and histopathology. The necropsy usually reveals

lesions in the esophagus and the aorta. Harrus et al. (1996), consider esophageal masses, scars and aorta aneurisms as pathognomonic of spirocercosis. The esophageal lesions are often granulomas. Not so often, however, sarcomas may occur. These are fibrosarcoma and osteosarcoma, and their incidence increases significantly in endemic areas (Bailey 1963, 1972; Ranen et al. 2004). Santos et al. (2004) report the presence of an aortic aneurism and esophageal granulomas in 11 stray dogs in Brasília and Gal et al. (2005) report a case of aortic thromboembolism. Here, we found nodules in the caudal border of the esophagus, as previously described by other authors (Bailey et al. 1963; Gosling and Gracia 1983). In accordance with Jones et al. (1997) the cross section of the nodules usually reveals a thick fibrotic wall, partially coated by epithelium, involving a cavity containing worms and yellow pus. The present work found nodules with firm consistency, very probably because of the presence of a fibrotic wall and one of the nodules contained a worm, corroborating with these authors observations.

The inertial agents such as helminths can lead to the formation of high substitution rate granulomas, with the presence of lymphocytes, macrophages with mitotic division, epithelioid cells and giant cells (Jones et al. 1997). These microscopic characteristics were not found and Bailey (1972) reports that there is usually a line that demarcates the reactive granuloma and the fibrous connective tissue, but this was not found. The presence of inflammatory cells and fibrosis occurred as characteristic, but not of demarcated mode. Alternatively, Jones et al. (1997) notes that pleomorphic cells in fibrosarcoma can vary from fuse-shaped and undifferentiated, with round or ovoid nucleus and frequent mitoses, to elongated cells, typical of immature tissue, and that the formation of spindle cell clusters is valuable information for the diagnosis of fibrosarcoma. This study identified little pleomorphism, without mitoses, but the feature of parallel cell formation (spindle-shaped) was compatible with the observations typical of fibrosarcoma. The empty cystic spaces probably correspond to the place where the parasite was located.

The present work is the first official report of the presence of canine spirocercosis in Guapimirim city. Following up to this, an epidemiological survey can be conducted in the city and other municipalities of the mountainous region. Conversely, this work can serve as a warning to clinical veterinary practitioners, where, in this probably

endemic region, the clinical presentation of food regurgitation may be an indicator of parasitism by *S. lupi* in dogs.

References

- Bailey WS (1963) Parasite and cancer: sarcoma in dogs associated with *Spirocerca lupi*. Ann N Y Acad Sci 108:890–923
- Bailey WS (1972) *Spirocerca lupi*: a continuing inquiry. J Parasitol 58:3–22
- Bailey WS, Cabrera DJ, Diamond DL (1963) Beetles of the family Scarabaeidae as intermediate hosts for *Spirocerca lupi*. J Parasitol 49:485–488
- Cabrera DJ, Bailey WS (1964) A modified stoll technique for detecting eggs of *Spirocerca lupi*. J Am Vet Med Assoc 145: 573–575
- Dvir E, Kirberger RM, Malleczek D (2001) Radiographic and computed tomographic changes and clinical presentation of spirocercosis in the dog. Vet Radiol Ultrasound 42:119–129
- Fox SM, Burns J, Hawkins J (1988) Spirocercosis in dogs. Compend Contin Educ Pract Vet 10:807–822
- Gal A, Kleinbart S, Aizenberg Z, Baneth G (2005) Aortic thromboembolism associated with *Spirocerca lupi* infection. Vet Parasitol 130:331–335
- Gosling E, Gracia A (1983) Diagnostico precoz y tratamiento de la spirocercosis (*Spirocerca lupi*). Vet Trop 8:57–69
- Harrus S, Harmelin A, Markovics A, Bark H (1996) *Spirocerca lupi* in the dog: aberrant migration. J Am Anim Hosp Assoc 32:125–130
- Jones TC, Hunt RD, King NW (1997) Disturbances of growth: aplasia to neoplasia. In: Jones TC, Hunt RD, King NW (eds) Veterinary pathology, 6th edn. Williams and Wilkins, Baltimore, pp 110–112
- Markovics A, Medinski B (1996) Improved diagnosis of low intensity *Spirocerca lupi* infection by the sugar flotation method. J Vet Diagn Invest 8:400–401
- Mense MG, Gardiner CH, Moeller RB, Partridge HL, Wilson S (1992) Chronic emesis caused by a nematode induced gastric nodule in a cat. J Am Anim Hosp Assoc 201:597–598
- Ranen E, Lavy E, Aizenberg I, Perl S, Harrus S (2004) Spirocercosis-associated esophageal sarcomas in dogs. A retrospective study of 17 cases (1997–2003). Vet Parasitol 119:209–221
- Ridgway RL, Suter PF (1979) Clinical and radiographic signs in primary and metastatic oesophageal neoplasms of the dog. J Am Vet Med Assoc 174:700–704
- Rudolphi CA (1819) Entozoorum synopsis cui accedunt mantissa duplex et indices locupletissimi. Sumptibus Augusti Rücker. Berolini, 811 p
- Santos ASO, Silveira LS, Lemos LS, Moreira L, Silva AC, Carvalho ECQ, Matos WR (2004) Aneurismas aórticos pela espirocercose canina em Brasília (relato de caso). Rev Bras Saúde Prod Anim 5:25–30
- van der Merwe LL, Kirberger RM, Clift S, Williams M, Keller N, Naidoo V (2008) *Spirocerca lupi* infection in the dog: a review. Vet J 176:294–309