CASE REPORT

EOSINOPHILIC APPENDICITIS CAUSED BY *SCHISTOSOMA JAPONICUM* : A CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract. Parasitic appendicitis is uncommon. The authors reviewed the pathology of 4,130 appendices resected over the past 10 years (2000 to 2009). Only one case of eosinophilic appendicitis caused by *Schistosoma japonicum* was identified. The overall prevalence of schistosomal appendicitis was 0.024%. The case was a 61-year-old woman who presented with right lower quadrant abdominal pain. She had been a farmer in Chumphon and Surat Thani Provinces, which are endemic for schistosomiasis in Thailand. Physical, laboratory and ultrasound examinations were suggestive of acute appendicitis. She underwent emergency appendectomy. Intraoperative findings revealed a ruptured appendix with a fecalith in the appendiceal lumen. The histopathologic diagnosis was suppurative eosinophilic appendicitis with schistosomal ova in the mucosa, submucosa, muscular layer and vascular lumens, identified as *S. japonicum* eggs. The patient was treated for the parasite with praziquantal. We briefly review the clinicopathologic features and pathogenesis of schistosomal appendicitis.

Key words: appendix, eosinophilic appendicitis, schistosomiasis, *Schistosoma japonicum*

INTRODUCTION

Parasitic infestations are one of the most important public health problems in tropical and subtropical regions of the world. However, parasitic appendicitis is an uncommon disease in human beings. *Enterobius vermicularis*, *Ascaris lumbricoides*, *Taenia* spp, *Opisthorchis viverrini*, and *Schistosoma* spp have been reported to be associated with parasitic appendicitis (Saechua and Saechua, 2006; Porncharoenpong, 2007; Karatepe *et al*, 2009). Schistosomiasis is a disease of water-borne trematode infestation caused by members of the superfamily Schistosomatoidea. *S. japonicum* and *S. mekongi* are two major species pathogenic to humans in Southeast Asia (Harinasuta, 1984). Schistosomiasis of the

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appendix was first reported by Turner in 1909. The most common Schistosomal species causing appendicitis are *S. haematobium* and *S. mansoni* (Satti *et al*, 1987; Adebamowo *et al*, 1991; Weber *et al*, 1998; Halkic *et al*, 2002; Adehossi and Parola, 2004; Madavo and Hurriez, 2006; Nandipati *et al*, 2008; Terada, 2009). To the authors' knowledge, there are five published reports of appendicitis caused by *S. japonicum* in the literature (Baughman *et al*, 1960; Ricosse *et al*, 1980; Palmieri *et al*, 1981; Moore and Smith, 1989; Lee *et al*, 1994). The prevalence of schistosoma appendicitis in Thailand is unknown. We report here a case report and the clinical, radiological, and histopathologic features and prevalence of schistosomal appendicitis during a ten year period (2000 to 2009) at a tertiary care university hospital in Thailand.

CASE REPORT

A 61-year-old woman presented to the emergency department with right lower quadrant abdominal pain for one day. She was born in Lang Suan District, Chumphon Province, southern Thailand and lived there for 22 years. After that she married and moved to Phra Saeng District in Surat Thani, southern Thailand and lived there for 26 years, where she worked as a farmer before coming to Bangkok and working as a housekeeper. Her underlying diseases were hypertension, dyslipidemia, and nodular goiter. Her symptoms started with colicky right lower quadrant abdominal pain and nausea for 1 day. On physical examination, she was afebrile, with a temperature of 36.8ºC, pulse rate of 80/minute, and a blood pressure of 168/85 mmHg. The abdominal examination showed voluntary guarding with active bowel sounds, marked tenderness without rebound tenderness of the right lower abdominal quadrant. The liver and spleen were not palpable. The rectal examination showed no tenderness. Relevant laboratory investigations included: hemoglobin 11.4 g/dl, hematocrit 36.6%, white blood cell count 13,300/mm^3 with 75% neutrophils, 4% bands, 16% lymphocytes, 4% monocytes and 1% eosinophils. Her urinary analysis and stool examination were normal. Ultrasound of the lower abdomen was done and exhibited an uncompressible blind-end tubular shaped structure in the right lower quadrant suggestive of acute appendicitis (Fig 1A, 1B). Emergency appendectomy was performed. A paracecal appendix was identified and it showed acute appendicitis with perforation. Postoperative antibiotics were ceftriaxone and metronidazole. The postoperative course was uneventful. The patient was discharged on the fourth postoperative day. The patient was treated with praziquantel for the schistosomal appendicitis.

The pathologic specimen consisted of a vermiform appendix measuring 6.5 cm long and 1.2 cm in average diameter. The serosa was coated with exudate (Fig 1C). There was a perforation 3.5 cm from the stump. The lumen contained a fecalith. The sections revealed acute inflammation with numerous eggs of *S. japonicum* in the lamina propria, submucosa, muscular layer and vascular lumens. The egg measured 60-80 by 50-60 μm with a minute lateral spine (Fig 1D, 1E, 1F). The eggs were surrounded by inflammatory cells composed of eosinophils, lymphocytes and macrophages. Some clusters of eggs were surrounded by fibrotic tissue. A few eggs were in the small vascular lumen. The muscular layer contained numerous neutrophils and eosinophils. The pathologic diagnosis was acute suppurative eosino-

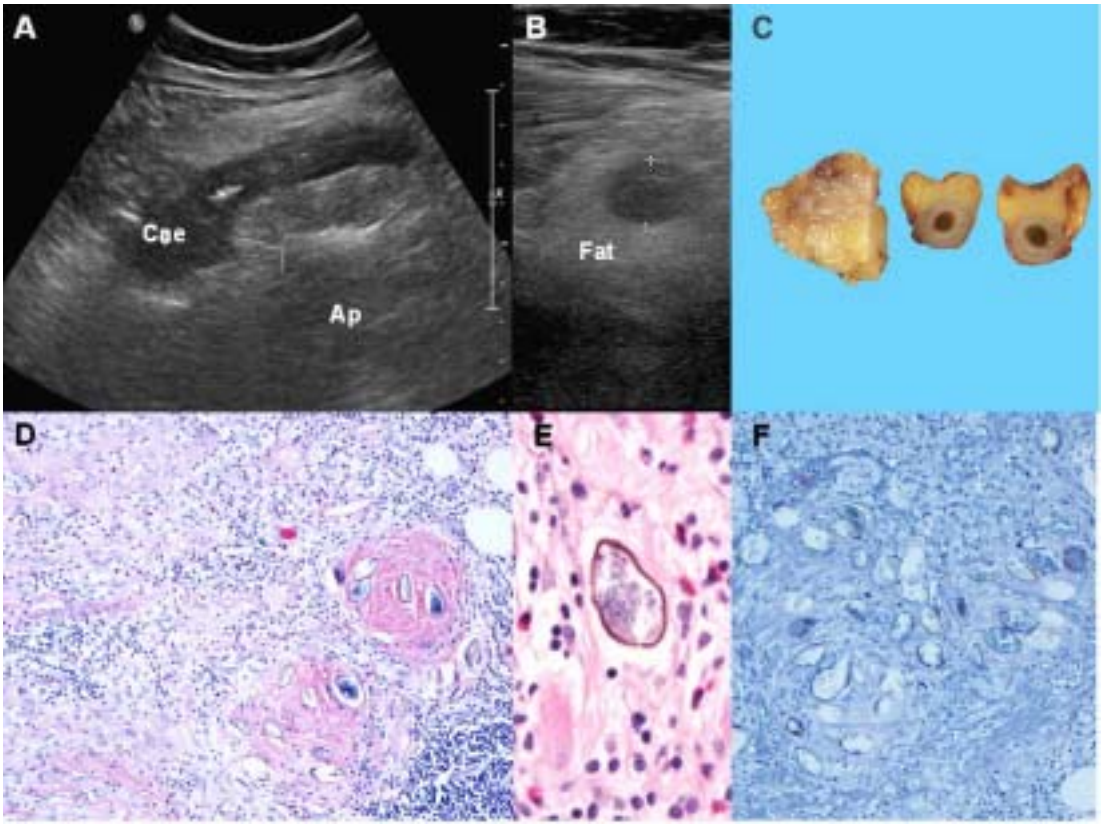

Fig 1–Ultrasound and pathologic findings of schistosomal appendicitis. A) oblique transabdominal ultrasound showing a blind-ending aperistaltic tubular structure with a thickened wall, corresponding with an acute inflamed appendix (Ap). "Cae" represents the cecum; B) transverse transabdominal ultrasound shows cross section of an enlarged, non-compressible inflamed appendix (12 mm in diameter), with surrounding periappendiceal fat inflammation (Fat); C) gross cross section of the inflamed vermiform appendix; D) deposition of schistosome eggs in the appendiceal wall with inflammatory cells composed of eosinophils, neutrophils, lymphocytes and macrophages. Some clusters of eggs are surrounded by fibrotic tissue. HE, x100; E) schistosomal eggs in the small vascular space. HE, x400; F) schistosomal eggs in the appendiceal wall. Ziehl-Neelsen stain, x100.

philic appendicitis with eggs of *S. japonicum*. There was also a fecalith in the lumen and an ulcer of the mucosa.

DISCUSSION

Schistosomiasis is a water-borne parasitic disease caused by blood flukes of the genus *Schistosoma*. It is endemic in Africa and Southeast Asia. In the Philippines, Indonesia and southern Thailand, the most common species are *S. japonicum* and *S. mekongi* (Harinasuta, 1984). *Schistosoma* parasites are an uncommon cause of appendicitis. In endemic areas, the incidence of schistosomal appendicitis has been reported to range from 1 to 4.2% (Weber *et al*, 1998; Badmos *et al*, 2006; Gali *et al*, 2006).

The most common *Schistosoma* species causing appendicitis are *S. haematobium* and *S. mansoni*.

Although *S. japonicum* is a primary cause of intestinal schistosomiasis, there are only five published reports of *S. japonicum* causing appendicitis (Baughman *et al*, 1960; Ricosse *et al*, 1980; Palmieri *et al*, 1981; Moore and Smith, 1989; Lee *et al*, 1994). *S. japonicum* live in the portal venous system and in mesenteric venules. Clinically significant intestinal schistosomiasis is caused by eggs deposited in the affected intestinal wall. The eggs of *S. japonicum* are generally deposited in the right colon. Petechial hemorrhages and granulomas in the lamina propria and submucosa are the most common lesions. *S. japonicum* infection includes acute and chronic phases. Acute phase symptoms can include serum sickness, fever, nausea, headache, an irritating cough and diarrhea, although most patients are asymptomatic. The chronic phase involves the liver, small intestine and large intestine, which may progress to liver cirrhosis, portal cirrhosis, polyp formation, strictures, fistulae, bowel perforation and appendicitis (Al-Waheeb *et al*, 2009). The pathogenesis of schistosomal appendicitis is described by two pathways. The first pathway is "granulomatous acute appendicitis" caused by an immunological granulomatous reaction to the newly laid ova with an eosinophilic response and tissue necrosis. The second pathway is "obstructive acute appendicitis" caused by chronic inflammation and fibrosis around dead ova, which stenoses the appendiceal lumen so feces can more easily lodge in the appendiceal lumen (Doudier *et al*, 2004).

In Nigeria, an endemic area, the incidence of schistosomiasis of the appendix is 2.3-4.2% (Satti *et al*, 1987; Adebamowo *et al*, 1991). Badmos *et al* (2006) suggested appendicitis could be caused by schistosomiasis but only two-thirds of cases of appendiceal schistosomiasis develop appendicitis. In Thailand, the incidence of eosinophilic appendicitis has been reported to vary from 0.32 to 2.63% (Saechua and Saechua, 2006; Porncharoenpong, 2007). Saechua and Saechua (2006) reported the incidence of eosinophilic appendicitis is 2.63% of all resected vermiform appendices, and in their study 20 out of 235 cases (8.51%) were caused by parasitic infestation. The etiologic parasites causing appendicitis were found to be: *E. vermicularis* (70-85%), *Taenia* spp (6.7-10%), and *O. viverrini* (5%) (Saechua and Saechua, 2006; Porncharoenpong, 2007). Eosinophilic appendicitis caused by *S. japonicum* has never been reported from Thailand.

With the case reported here, the patient had no symptoms of schistosomal infection prior to presenting with symptoms of infection. The source of the infection may have been from her hometown in southern Thailand where she used to work the ground as a farmer. The pathology of the case reported here showed eosinophilic suppurative appendicitis with *S. japonicum* eggs in the mucosa, submucosa, muscular layer, and microcirculation. Chronic inflammation and fibrosis around the ova were identified. These findings are not seen in acute appendicitis caused by primary obstruction with secondary bacterial infection. What is seen with typical appendicitis is neutrophilic infiltration in the appendiceal wall. Tissue eosinophilia in the appendix provides evidence for parasitic infestation. The diagnosis of schistosomiasis in endemic areas of Thailand is usually made by finding schistosome eggs in feces. *S. japonicum* eggs measure 70-100 by 50-65 μm in diameter, are oval to round in shape with a subterminal

spine, and have a hematoxyphilic shell (Cheever and Neafie, 2000). Standard and modified Ziehl-Neelsen stains can be useful in identifying schistosome eggs in tissue sections. The shell and the spine of *S. japonicum* are acid-fast, whereas only the spine of *S. haematobium* is acid-fast (Cheever and Neafie, 2000). However, the shell and spine sometimes fail to stain as expected.

Schistosomiasis is a serious parasitic infestation with a high morbidity rate. Early diagnosis and prompt treatment are essential. It has a wide spectrum of clinical manifestations. Current advances in immigration in a borderless world have resulted in an increasing population at risk for parasitic infection. Recent technology has not improved the sensitivity and overall accuracy of clinical diagnosis of parasitic infestation. Therefore, routine histopathologcal examination of the appendix is of value for identifying unsuspected conditions and may reveal unexpected findings of clinical importance necessary for the diagnosis, treatment and prevention of the infectious disease.

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