

Oesophagostomiasis in Man: report of the first Malaysian case with emphasis on its pathology

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Abstract

This paper describes the first Malaysian case of oesophagostomiasis. The patient was an 8-year-old Malay boy who presented to Ipoh General Hospital, Perak with abdominal pain and weight loss. Examination revealed multiple cavitated pseudotumours of the colon. A hemicolectomy was performed. Examination of the lesions revealed Oesophagostomum worms in the necrotic centres. The differential diagnoses and the pathology caused by Oesophagostomum are discussed. A brief review of the available literature is presented.

Key words: Oesophagostomum, colon, pseudotumour, subcutaneous nodule.

INTRODUCTION

Nematode worms of the genus *Oesophagostomum*, superfamily Strongiloides, are not well recognised for producing symptomatic disease in man but commonly invades apes, monkeys and herbivores.

Up to the present, only about 70 cases of human infection with Oesophagotomum have been recorded.' Most of these cases were from Africa^{2,3,4,5} while a few have been reported from Indonesia⁶ and South America.' In most of the cases, the lesions were described as inflammatory nodules, pseudotumours or abscesses.

The aim of this paper is to present the first case from Malaysia, to compare it with cases described elsewhere and to create an awareness of the infection in this country.

CASE REPORT

The patient was an eight-year-old Malay boy from Grik, Perak, who was admitted to the Ipoh General Hospital with a history of abdominal pain and loss of appetite of 2 days duration. There was no vomiting or diarrhoea during that period. The abdominal pain was colicky in nature. It started over the pen-umbilical region and later involved the whole abdomen. This was associated with abdominal distension and intermittent low grade fever. The lungs were clear during examination. No lymphadenopathy or organomegaly was detected. Tenderness and guarding were present over the whole abdomen and there was no visible peristalsis or bowel sounds. Blood examination did not reveal any significant electrolyte

disturbance. The serum amylase and protein levels were within normal limits. The haemoglobin level was 10.8 gm per 100 ml. The white blood cell count was 15,700 per cmm, but there was no eosinophilia. Urine examination revealed no abnormality.

Based on the above findings, a presumptive diagnosis of acute peritonitis due to ruptured appendix was made. Laparotomy via a right pararectal incision was performed. Multiple pseudotumours over the ante-mesenteric border of the large intestine, extending from the caecum to descending colon were found. The mesenteric and paracolic lymph nodes were significantly enlarged. Many worms were found with pus and free fluid in the peritoneal cavity, together with numerous perforations along the large intestine. An extended hemicolectomy was performed and the terminal ileum was anastomosed to the proximal part of the descending colon. The patient's post-operative course was uneventful. A few weeks later, he developed a nodular swelling in the anterior abdominal wall over the epigastric region. It was subsequently excised. The patient was discharged a few days after the operation.

The excised specimens were fixed in 10% formalin and representative samples of the lesion submitted for histological examination. Special stains such as Van Gieson were used when necessary.

PATHOLOGY:

Hemicolectomy specimen

The resected large bowel consisted of the caecum, and the ascending, transverse and proximal part

of the descending colon, altogether measuring 35 cm in length.

The surface of the colon was studded with 20 well-defined nodules between the external muscular layer and the peritoneal covering. The surfaces of the nodules were generally smooth although a few appeared rough and covered with inflammatory exudate and adhesions. Occasionally, perforations of the nodules were noted. The nodules were generally small and round to oval in shape, varying between 3 mm-15 mm in size, and elevated as much as 5 mm - 7 mm above the surface of the bowel.

The nodules were opaque and greyish-black in colour. The locations of the nodules varied. Some were found along the mesenteric attachment and others at the ante-mesenteric border. The nodules were found either 2 cm-4 cm apart or in small groups of 2-4 oval tumours about 2 cm from one another. Occasionally, isolated ones were seen.

Many of the small nodules were hard. The larger ones were soft, compressible and pitted freely on pressure. The nodules appeared to be firmly attached to the wall of the colon. On sectioning, some of these nodules revealed small worms lying in semisolid dark brown material. The worms were either coiled or stretched out. Many of the paracolic lymph nodes were enlarged, about 0.5 cm-1.0 cm in the largest dimension.

On opening the colon, some fifty nodules were situated in the walls (Fig. 1), the majority of them causing more distinct bulging of the mucous membrane than those situated on the external surface of the intestine. Examination showed narrowing of the lumen of the gut, so much so that it was difficult to allow the passage of the tip of a little finger. Most of the nodules were oval in shape, measuring 5 mm by 5 mm up to 10 mm by 25 mm with average bulging of the mucosa from 1 mm-10 mm.

The nodules in the submucosa generally lay transversely or diagonally. A few were longitudinally to the bowel axis. They were soft and compressible. Many were spongy, and moderate pressure sufficed to rupture their fragile walls. In cross sections of the colon, the nodules appeared to be situated variously in the subperitoneal and submucosal fascia, and between the internal and external muscular coats, causing a honeycomb appearance. On sectioning, the nodules revealed central cavities containing dark-brownish semi-solid material in which worms were embedded, enclosed by thick walls with smooth lining (Fig. 2). No calcareous deposits were found.

Microscopical examination

Histology revealed pyogenic colitis with peritonitis. The nodules were located in the subperitoneal, intramuscular and submucosal regions, often compressing the muscularis mucosae and internal muscular coat (Figs. 3 and 4). The walls of the nodules were composed of connective tissue and granulation tissue infiltrated by macrophages and foreign body giant cells. The contents consisted of degenerated erythrocytes, necrotic tissue debris, neutrophils, lymphocytes, plasma cells, pigment-laden macrophages, multinucleated giant cells and clumps of granular pigment. Few eosinophils were noted. The central granular debris contained numerous Charcot-Layden crystals and calcified oval and spherical bodies (Fig. 5). In some of the sections, parts of the worms were seen (Fig. 6). The enlarged paracolic lymph nodes showed reactive hyperplasia and prominent germinal centres. Scanty eosinophilia was observed.

Anterior abdominal wall nodule

Macroscopically, the specimen was almost spherical measuring 3.5 cm x 3.5 cm x 3 cm. It was firm and greyish-yellow in colour. On sectioning, a central cavity filled with yellowish-green pus-like material was seen. No worm was identified.

Microscopically, the wall consisted of thickened fibrous tissue. The contents consisted of necrotic tissue, inflammatory exudate and numerous calcified bodies at the inner wall, presumably due to calcified dead worms.

Parasitology

20 worms obtained from the peritoneal cavity and a few from nodules of the resected colon were studied by the parasitologist using light microscopy and scanning electron microscopy (SEM).

The worms were all immature adults, creamy white in colour when preserved in 5% formalin. As characteristic of the genus, they were distinguished by a cephalic vesicle separated from the body by a transverse ventral groove. The mouth was directed straight forward and surrounded by two lateral and four submedian head papillae. The copulatory bursa, seen in the tail of the male worms, was characterised by the cleft ventral rays, the proximally fused mediolateral and posterolateral and the branching of the external dorsal and dorsal. The tail of the female was sharp pointed, with the vulva just anterior to the anus.

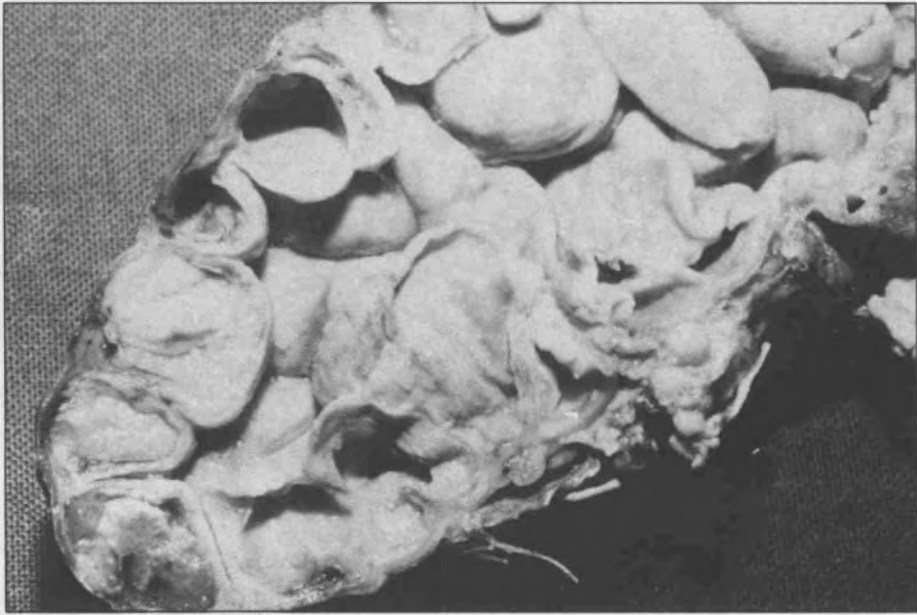


FIG. 1: Interior surface of colectomy specimen showing extensive involvement of the caecum by helminthoma.



FIG. 3: Magnified view of the caecum. Cut-section through a helminthoma reveals a worm (arrow). The walls of the cysts consist of fibrous tissue and the central cavities are filled with yellowish pus-like material.



FIG.3: Microphotograph of the wall of the colon showing abscesses in the submucosa, muscle and subserosa.

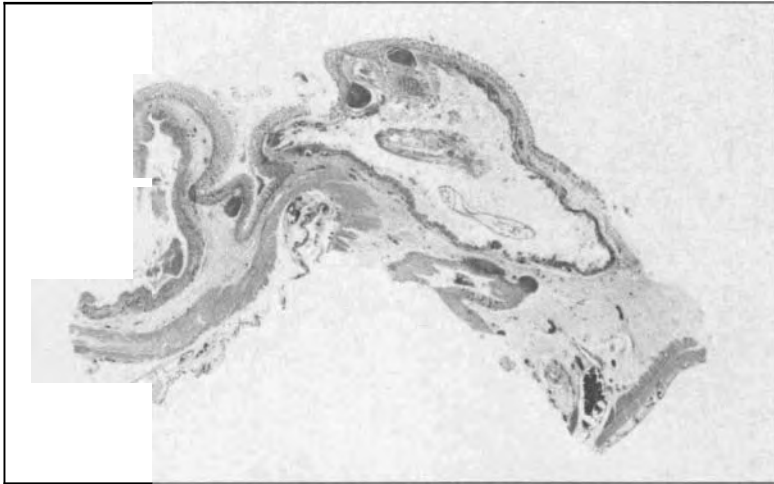


FIG. 4: Microphotograph showing that the central cavity of a helminthoma contains necrotic tissue and the cross-section of a worm.

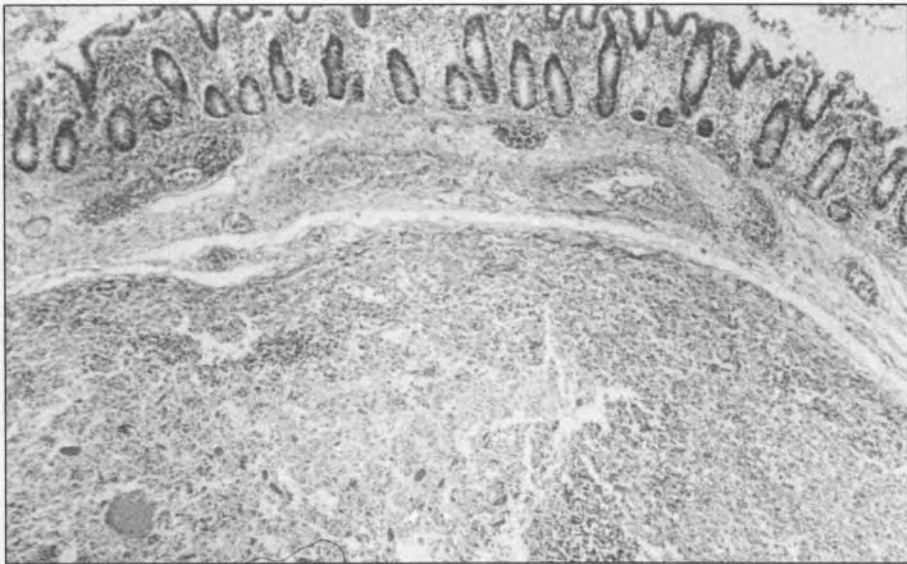


FIG. 5: Abscess cavity surrounded by fibrous tissue and acute and chronic inflammatory cells, macrophages and calcified bodies.



FIG. 6: Cross-section of a worm in an abscess cavity.

Female worms examined averaged 10.4 mm (range = 0.8 mm - 11.8 mm) long and 0.5 mm across the mid-body. They were non-gravid with the vulva situated 405 µm (381 µm - 530 µm) and the anus 202 µm (159 µm - 223 µm) from the tip of the tail.

The only reliable character that appeared to be of value in identification of the worms to species laid in the corona radiata. Both male and female worms have 10 sharply pointed leaf-like elements in the external leaf crown. The internal leaf crown is in the form of 10 pairs of much smaller, similarly-shaped elements, with each pair lying in between the elements of the external leaf but located well beneath the base. The worms seemed identical to those described as *O. apistomum* (*O. aculeatum*) from Indonesia.⁸

DISCUSSION

Oesophagostomiasis is a common pathogenic parasitic infection of pigs, sheep, goat, cattle, apes and monkeys and has been found, on rare occasions, in man. As the ova of the worms are indistinguishable from those of hookworms, human cases may have been missed and may be more common than suspected. Human infection has only been reported from areas where monkeys abound such as East Africa, South America and Indonesia. Both sexes, children and adults have been affected.

It is not possible to deduce a characteristic clinical picture of the infection from available case reports. The clinical course appears to vary considerably. Elmes and McAdam noted no uniform clinical picture besides signs and symptoms of intra-abdominal inflammation.⁹

An abscess may rupture into the peritoneum to produce peritonitis. Oesophagostomiasis has also been documented as a cause of diarrhoea and peritonitis in Brazil. Differential diagnoses have included appendicitis, diverticulitis, Crohn's disease, carcinoma, amoebiasis and tuberculosis.¹⁰ Awareness of the condition may have led to a diagnosis in the present case but it is believed that many cases may have gone unrecognised.

The clinical presentation of helminthomata has been described by Welchman¹¹ as occurring in four ways:

- a) abscess of the abdominal wall in childhood with a short history of pain and fever.
- b) a picture suggestive of intussusception with fever, an abdominal tumour and pain. The duration may be anything from three days to three weeks.
- c) similar to appendicitis in adults where there

is a history of one to five days of abdominal pain and tenderness or a tender mass in the right iliac fossa.

- d) miscellaneous abdominal lumps in adult patients.

In another study, none of the patients had a blood picture characterised by marked eosinophilia though slight eosinophilia was present in 2 out of 9 patients.⁵ Examination of the stools for eggs was unrewarding as they were extremely difficult to differentiate from other nematode ova.⁵ In general, the diagnosis was often only made at laparotomy.¹²

Histological examination of oesophagostoma lesions revealed extensive oedema, necrosis and chronic inflammation and fibrosis involving the submucosa, muscularis and pericolonic fatty tissues. The inflammatory reaction had no specific features although there were usually eosinophils, plasma cells, lymphocytes and macrophages within the granulation tissue. None of these features were specific in themselves, and all might be seen in diverticulitis. The ileo-caecal region was most frequently involved followed by the transverse colon, ileum sigmoid colon and mesentery.⁵

The term "helminthoma" was first used by Elmes and McAdam,³ to designate a tumour-like inflammatory swelling of the bowel following penetration of the wall by a nematode worm. Since the clinical presentation was variable, misdiagnosis was frequent and the pathologist may be baffled by appearances that range from inflammatory to neoplastic. Carcinoma has been frequently suspected⁵ and unnecessary radical surgery carried out.

It is particularly important that pathologists be aware of the existence of this condition as they are the usual persons to make the diagnosis unless the surgeon has been fortunate enough to find the worms. In cases where worms are found, a definite diagnosis is ensured. If none are found, these cases can be confused with tuberculosis, the histological appearances of which they may superficially resemble. Anthony and McAdam⁵ studied 34 cases, and classified the lesion into classes I, II and III representing stages in the inflammatory process.

Class I represented the acute lesion. This is characterized by an ill-defined abscess or track filled with a fibrinous exudate in which inflammatory cells are scanty. Worms are likely to be found although the whole of the mass may need to be sectioned in the search. In Class II when the abscess or track becomes walled off by

a layer of macrophages, epithelioid cells, fibroblasts and the contents become cheesy. No recognisable worm is seen and granular calcification results from dead worms that are being reabsorbed. Class III is the chronic fibrotic lesion with epithelioid and giant cell granulomata and variable eosinophilic leucocytic infiltration.

In all but one of the reported cases, oesophagostomata were confined to the intestine. Chabaud and Lariviere¹³ reported a case producing a subcutaneous tumour in the abdominal wall of a child. The present case appears to be the second reported in which the worms have produced lesions in the gastrointestinal tract as well as caused a subcutaneous nodule. It seems likely that this nodule was caused by intraperitoneal infection spreading to the anterior abdominal wall from the primary infection in the intestine.

Little evidence is currently available on how infection with esophagostomes is acquired.¹⁴ Infection with first and second stage larvae is thought to occur perorally and is followed by a third moult, usually in the caecum. The third stage larva is histotropic and migrates into the submucosa, usually of the large bowel, resulting in the formation of nodules. Here the larva matures further prior to returning to the bowel lumen where full maturity is achieved. The empty nodule then shrinks and calcifies.¹⁵ In certain cases this process is interrupted and the worms remain within the nodules where they are able to survive for several months.¹²

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