CASE REPORT

Computed Tomography Detection of Schistosomal Appendicitis

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ABSTRACT
Schistosomiasis is the second most prevalent parasitic disease worldwide, which is rarely reported in developed countries like Hong Kong. We report a case of schistosomal appendicitis in a 79-year-old male presenting with right lower abdominal pain. Computed tomography of the abdomen and pelvis showed dilated appendix with adjacent stranding, suggestive of acute appendicitis. Long-segment circumferential interrupted mural calcification was also seen along the large bowel and appendix, together with capsular calcification extending towards the centre of liver with turtleshell appearance, highly suspicious of prior schistosomiasis infection. Subsequent histological study of the appendicectomy specimen revealed transmural suppuration along the appendix with numerous calcified Schistosoma ova in the appendiceal wall, compatible with schistosomal appendicitis. Having discussed the incidence and pathogenesis of schistosomal appendicitis, we stress the importance of considering prior schistosomal infestation as a cause of acute appendicitis, especially in patients who have emigrated or are travelling from endemic areas.

Key Words: Appendicitis; Diagnosis; Parasites; Schistosomiasis; Tomography, X-ray computed

中文摘要
利用電腦斷層掃描檢測血吸蟲闌尾炎
鄧峻樺、宋咸東、林聞華、鄭志成
血吸蟲病是全球第二種最常見的寄生蟲病，發達國家如香港很少有類似的病例報導。本文報告一名症狀為右下腹痛的79歲男性血吸蟲性闌尾炎病例。病人的腹部和盆腔電腦斷層掃描顯示闌尾擴張伴周圍脂肪條紋徵，提示急性闌尾炎。沿大腸和闌尾可見長段環狀非連續性壁鈣化，伴囊狀鈣化延伸至呈龜甲外觀的肝臟的中心位置，高度懷疑原發性血吸蟲感染。闌尾切除術的病理標本證實為透壁性化膿性闌尾炎伴闌尾壁上多個鈣化的血吸蟲卵，符合血吸蟲闌尾炎的診斷。本文討論血吸蟲性闌尾炎的發病率和發病機制，強調考慮原發性血吸蟲感染為急性闌尾炎的病因的重要性，特別是對於曾移民或到血吸蟲病流行地區旅行的病人。

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INTRODUCTION
Schistosomiasis is the second most prevalent parasitic disease worldwide. More than 200 million people are infected, in which 120 million are symptomatic, and 20 million suffer from severe disease. It is most common in Africa, South America and Asia, but rarely reported in developed countries. An estimated 85% of all infected people are from African countries. Due to extensive emigration and globalisation, the incidence is rising in developed countries.

CASE REPORT
A 79-year-old male presented to the local emergency department in Hong Kong in November 2013, with colicky right lower quadrant abdominal pain for 3 days. He had underlying diabetes mellitus and hypertension. On physical examination, he was afebrile, with a blood pressure of 110/58 mm Hg and pulse rate of 98 beats/min. Abdominal examination showed tenderness and voluntary guarding without rebound tenderness in the right lower abdomen. Relevant laboratory investigations included haemoglobin level of 131 g/L and white blood cell count of 15.22 x 10^9/L. Urgent contrast computed tomography (CT) of the abdomen and pelvis was performed which showed circumferential wall thickening of the dilated appendix with adjacent stranding and prominent lymph nodes. A 5-mm wall defect was noted at the base of the appendix (Figure 1).

Figure 1. (a) A coronal image of contrast computed tomography (CT) shows dilated appendix with adjacent abscess formation. Focal wall defect (arrow) is seen at the base of the appendix. Mural calcification is seen in the ascending colon (arrowheads). (b) Sagittal and (c) axial images of pre-contrast CT show circumferential interrupted mural calcification (arrows) of the appendix and sigmoid colon. (d) An axial image of pre-contrast CT shows capsular calcification extending towards the centre of liver with turtleback appearance, suggestive of prior schistosomiasis infection.
Features were suggestive of acute appendicitis with perforation. Long-segment circumferential interrupted mural calcification was seen along the large bowel and appendix (Figures 1b–c). Capsular calcification extending towards the centre of liver with turtleback appearance was noted (Figure 1d), giving rise to a suspicion of prior schistosomiasis infection. Liver cirrhosis with splenomegaly was also seen.

Laparoscopic appendicectomy was performed, which showed ruptured appendicitis with retrocaecal abscess. Faeculent material drain output was noted on day 3 after surgery. Surgical exploration showed gangrenous caecum with rupture and, therefore, a limited right hemicolectomy was performed. Histological study by light microscopy revealed transmural supplicative inflammation of the appendix with haemorrhage and extensive necrosis. Large number of calcified Schistosoma ova were found in appendiceal, ileal and colonic wall, predominantly in the submucosa and focally extending to the mucosa, accompanied by increased chronic inflammatory infiltrate in the mucosa (Figure 2). Stool for ova and cysts were negative on two separate examinations. The patient lived in Shanghai since birth, and he came to Hong Kong at 15 years of age. He had a history of swimming in a freshwater lake in Hong Kong. The patient was treated with praziquantel and discharged after 23 days.

DISCUSSION
Schistosomiasis is a tropical parasitic disease caused by blood-dwelling fluke worms of the genus Schistosoma. The main schistosomes infecting human beings are Schistosoma haematobium, Schistosoma japonicum, and Schistosoma mansoni. Appendicitis is one of the most common causes of surgical abdomen, with various unusual presentations.

Schistosomiasis as a cause of acute appendicitis is reported in 0.02% to 6.3% of patients.1 Reports from developed countries, such as Japan, reviewing 311 appendectomy specimens revealed an incidence of 0.32%.2 Schistosomiasis was first described by Theodor Bilharz, and reported as a cause of urinary schistosomiasis. Schistosomal appendicitis was first described by Turner in 1909 as reported by Hodasi.3

Two pathways describe the pathogenesis of schistosomal appendicitis. The first mechanism is granulomatous acute appendicitis, described as an immunological granulomatous reaction to newly deposited ova, with subsequent tissue necrosis and eosinophilia. It may occur early in the infection, i.e. within weeks. The second mechanism is obstructive acute appendicitis, caused by chronic inflammation and fibrosis around the dead ova, leading to obstruction of the appendiceal lumen, increasing the risk of infection from faecal contaminants. This may occur in the late stage of infection after several months or years.4,5

Adult Schistosoma worms often reside in the tributaries

[Figure 2. Numerous blue-black, amorphous, calcified Schistosoma ova in the submucosa of the appendix: original magnification (a) x 100 and (b) x 400 (H&E).]
of portal vein, and produce eggs in the small intestinal and mesenteric veins. About 25% of the eggs penetrate the intestinal wall and leave the human body through faeces. The remaining eggs either flow upstream through the portal vein or stay in the intestinal wall. They cause inflammatory reaction and fibrosis. Subsequently, they die and are calcified, mostly in the intestinal wall and liver. They could also deposit in the submucosa of the appendix and cause calcification, which predisposes to appendicitis by the mechanisms described above.

With an increasing number of CT abdominal scans being performed in daily practice, there would be more cases of incidental finding of large bowel mural calcification in asymptomatic patients. Possibility of schistosomiasis could be emphasised on CT report, to alert clinicians to further look for evidence of infestation such as ova in the stool or urine and ultrasonographic images of liver, and help in timely institution of subsequent eradication treatment. The colonic calcification from schistosomiasis should be distinguished from other causes such as sclerosing encapsulating peritonitis, phlebosclerotic chronic ischaemic colitis, mucinous adenocarcinoma, and leiomyosarcoma. The colonic calcification from schistosomiasis typically involves the inferior mesenteric vein territory, as the schistosomal worms live mainly in the inferior mesenteric vein.8 Besides mural calcification, sclerosing encapsulating peritonitis may show dilated and tethered small bowel encased within a thickened peritoneum.9 Phlebosclerotic chronic ischaemic colitis usually involves the right-sided colon compared with distal colonic involvement in schistosomiasis, with characteristic mesenteric venous calcification.9a Mucinous adenocarcinoma often involves the short segment of the colon and may cause bowel obstruction due to severe luminal narrowing. In leiomyosarcoma, CT scans typically reveal colonic wall thickening with bulky mural masses with calcification, exophytic growth, heterogeneous enhancement, and cystic degeneration.11

Schistosomal appendicitis is a rare disease in Hong Kong and developed countries, with characteristic mural calcification of the infested appendix on CT scan. It should be considered a cause of acute appendicitis, especially in patients who have emigrated or are travelling from endemic areas. It is essential to include schistosomiasis as a differential for colonic mural calcification in asymptomatic patients to alert clinicians for further evaluation and institution of subsequent eradication treatment.

REFERENCES