CASE REPORT

A RARE PATHOLOGICAL TRIAD UNMASKED BY TYPHOID ENTERITIS: REPORT OF A CASE OF ACUTE APPENDICITIS, APPENDICEAL SCHISTOSOMIASIS AND VILLOUS ADENOMA OF THE APPENDIX.

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ABSTRACT

A villous adenoma of the appendix is an extremely rare tumour of the appendix even rarer is its occurrence on schistosomal granulomatous appendicitis. It is therefore usually an incidental finding during appendicectomy. Most series on appendiceal tumours accounts for less than 7% as villous adenomas. The index case histologically shows acute appendicitis, schistosomal granulomata and a villous adenoma. There may be an aetiological relationship between schistosomiasis and the villous adenoma of the appendix in this case since schistosoma related colonic squamous cell carcinomas have been observed in endemic arrears. Treatment is limited to appendicectomy and praziquantel therapy but post-operative colonoscopy is recommended due to the risk of colonic schistosomiasis and malignancy in patients with appendiceal neoplasms. We therefore report this rare pathological triad.

KEYWORDS : Appendicitis, Schistosomiasis, Villous Adenoma, Typhoid Enteritis.

INTRODUCTION

The appendix in humans is a rudimentary structure with no known function arising from the medial wall of the caecum and its mucosal lining is similar to that of the large bowel[1]. Acute appendicitis is predominantly a disease of the western world while chronic inflammation secondary to parasitic infestations is commoner in the third world[1]. Schistosomal appendicitis forms about 2% to 3% of appendectomies in some studies[2]. Tumours of the appendix are rare and are usually found incidentally during surgery for acute appendicitis[3]. Connor *et al* found 74 appendiceal tumours in 7,970 appendectomies and only 7% were villous adenomas[4]. Villous adenoma progressing to adenocarcinoma of the appendix has been reported previously[5]. Association of schistosomal colitis with colonic carcinoma has also been previously described[6]. However, schistosomal appendicitis associated with appendiceal villous adenoma has not been reported to the best of our knowledge. We present here a rare case of acute appendicitis on chronic schistosomiasis and appendiceal villous adenoma unmasked by typhoid enteritis.

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CASE REPORT

A 13 year old boy was referred to our teaching hospital by a private clinic with two weeks history of fever and colicky abdominal pain and subsequently abdominal distension and melena stool two days prior to presentation. He was pale and received two units of blood at the referral clinic. Examination revealed a male teenager in painful distress and generalized abdominal tenderness. Ultrasound scan revealed distended bowel loops with intraluminal fluid.

There was a small collection of fluid in the recto-vesical pouch but no significant ascites. Blood chemistry revealed a creatinine level of 132µmol/L with all other parameters remaining within normal limits. The PCV was 22%. Blood culture was not done. An impression of typhoid enteritis with perforation was made. The patient had laparotomy that revealed a 0.4 cm ileal perforation at the anti-mesenteric border and 30 cm from the ileo-caecal valve which was refreshed and closed. Further examination revealed an inflamed appendix which was also removed. The patient was given antibiotic cover and did very well post-operatively.

Histological examination of the appendix revealed transmural acute appendicitis, transmural deposits of embryonated and calcified schistosome ova and sessile villous adenoma (Figure 1, Figure 2 and Figure 3 respectively).

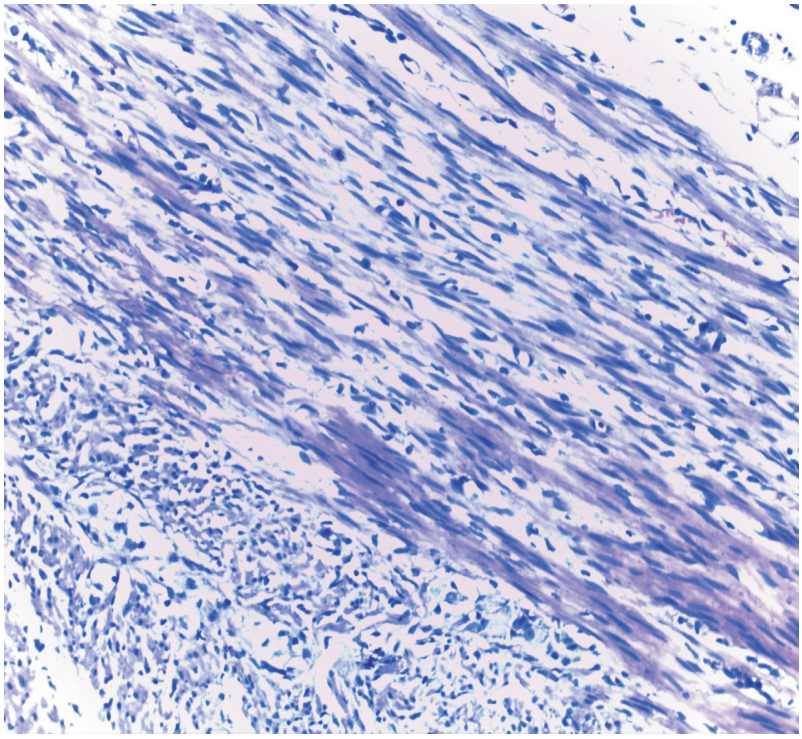

Figure 1: Transmural acute inflammation composed of polymorphonuclear leukocytes and eosinophils (H&E x 40)

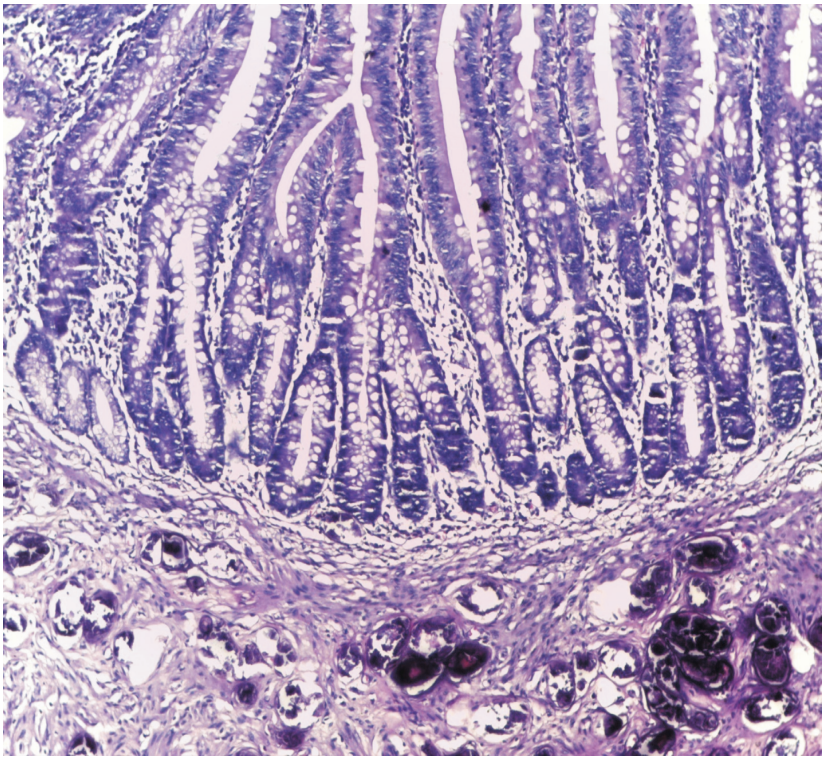

Figure 2: Sessile villous adenoma directly overlying embryonated and calcified schistosome ova (H&E x 40).

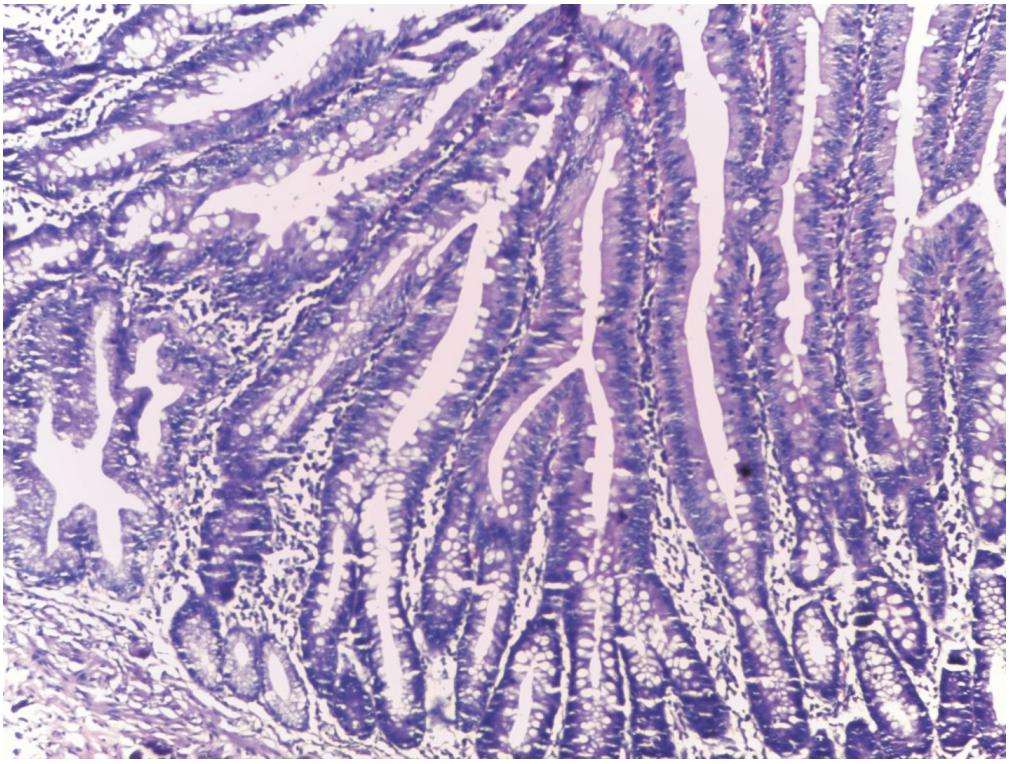

Figure 3: Sessile villous adenoma (H&E x 40)

DISCUSSION

Acute appendicitis is said to be predominantly a disease of the western world especially USA and Great Britain and is less common amongst Asians and Africans[1]. Acute appendicitis results from luminal obstruction usually from a faecolith or lymphoid hyperplasia[7]. About 4.2% of all cases of acute appendicitis show involvement of the appendix by schistosomiasis characterized by oviposition in some schistosomiasis endemic areas[8]. This leads to sub-mucosal fibrosis, narrowing of the lumen, obstruction and subsequent inflammation[8]. Schistosomiasis is known to be associated with the development of carcinoma of the bladder but gastrointestinal cancers associated with schistosomiasis have been found to be far less common[9]. The International Agency for Research on Cancer (1994) considered Schistosoma haematobium as carcinogenic to humans (group 1 carcinogen) and Schistosoma Japonicum as possibly carcinogenic to humans (group 2B carcinogen)[9]. Association has been established between goblet cell carcinoid tumour of the appendix and schistosomiasis of the appendix[9] while another study shows colorectal carcinomas in 32 patients associated with schistosomiasis[6]. Cases of appendicitis associated with villous adenoma so far reported did not include schistosomiasis. Our patient although presenting because of typhoid enteritis/perforation had appendicitis associated with schistosomiasis and villous adenoma either of which could have led to acute appendicitis. The typhoid perforation was closed surgically and the appendix was removed. The most important point to be noted in this association is the possible aetiological link between schistosomiasis and villous adenoma. This is because of the colorectal cancer cases observed in some schistosome-related enteropathies[6]. However, a cause and effect relationship has not been fully established between the two conditions. This possibility is high in our patient as the deposited ova are directly below the adenoma of the appendix. Since the appendiceal oviposition is likely to have come from the recto-sigmoid colon further monitoring with recto-sigmoidoscopy becomes mandatory while treating the patient with praziquantel. The possibility of synchronous colonic adenoma exists in these types of patients as villous adenomas are known pre-cursors of colorectal cancers and carcinogenesis associated with helminthes infections such as schistosomiasis is a complex process but chronic inflammation amongst other mechanisms appears to play a central role[6]. Chronic inflammation can generate nitrogen species and free radicals which can damage and oxidize DNA and lead to genetic instabilities which may subsequently evolve into atypical epithelial hyperplasia through proliferative polyps and finally neoplastic transfortion[6]. This is a feared complication in our patient however, removal of the appendix may have cured our patient as the adenoma was away from the resection margin and close follow-up since surgery did not reveal new complaints. Other studies on appendiceal adenomas made similar observations post-operatively[7].

CONCLUSION

This is the first case report to the best of our knowledge of this triad of acute appendicitis on appendiceal schistomiasis and villous adenoma of the appendix and provides a brief review of the literature. There is need for every appendix removed to be examined histologically. There is need to do colonoscopy on these type of patients to exclude synchronous or metachronous colonic adenomas to avoid the danger of malignant transformation.

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