CLINCAL CASE

BENIGN MASQUERADE – A RARE CASE OF COLONIC MALIGNANCY LURKING BEHIND AMEBIC COLITIS

Dharamanjai Kumar Sharma[1], Hina Sharma[2]

[1]Department of Surgery, Rabindra Nath Tagore Medical College, Udaipur, Rajasthan, India
[2]Department of Anatomy, Geetanjali Medical College & Hospital, Udaipur, Rajasthan, India

Corresponding author: Dharamanjai Kumar Sharma
E-mail: drdksurg@gmail.com

Abstract

A 45-year-old female patient presented with intestinal obstruction due to rectal stricture and associated radiologic findings and histopathology indicative of invasive amoebiasis. Suspicion of neoplasia arose due to incomplete response to anti-amoebic therapy which could only be proved after repeated biopsies. Amoebic colitis has been reported to masquerade as colorectal malignancy. In contrast, we herein present a case of rectal malignancy presenting primarily as amoebic colitis, creating considerable difficulty in diagnosis and management. This "benign masquerade" in which a malignant disease mimics or is masked by a benign pathology is particularly problematic since it lets the malignant pathology progress. A failure of complete resolution of amoebic lesions with anti-amoebic therapy should be considered a red flag and possibility of a more sinister coexisting pathology must be excluded in such cases. The patient had generalized invasive colonic amebiasis which is extremely rare. The unique radiologic appearance of generalized invasive colonic amebiasis is illustrated.

Keywords*: invasive amoebiasis, amoeboma, colitis, malignant masquerade*

Introduction

The clinical spectrum of amebiasis includes a variety of distinct forms (e.g., acute colitis, amoeboma, toxic megacolon, fulminant necrotizing colitis, colonic stricture, etc.) which closely resemble manifestations of other diseases. With ease of travel, amebiasis is now a truly global disease, making it an important differential diagnosis of colorectal diseases. The diagnostic dilemma is compounded if amebiasis happens to co-exist with some other colorectal pathology, especially malignancy. Only a small proportion of patients with amebiasis develop its invasive variant [1] and it is even rarer to find it co-existing with colorectal malignancy [2]. However, it is important to recognize this combination because rapid deterioration can occur if the diagnosis and treatment of either pathology are delayed or inappropriate.

Invasive colorectal amebiasis is usually localized and truly generalized invasive amebiasis of colorectum is extremely rare. We present a unique case with both generalized and localized invasive colorectal amebiasis co-existent with colorectal malignancy illustrating difficulties in diagnosis and management in such a scenario. This problem is more likely in areas endemic to amebiasis since colorectal complaints are more likely to be attributed to it compared to non-endemic regions. We discuss the unique

features of our case as well as management issues arising from this.

Case presentation

A 45-year-old female patient presented with acute intestinal obstruction of 3 days' duration. On digital rectal examination, a tight circumferential stricture without any palpable growth was found in distal rectum. Prior to this, the patient had complaints of mild abdominal distension and pain for 4 months. At that time, abdominal ultrasound had shown marked mural thickening in descending colon up to rectum with obliteration of the lumen and distension of proximal segments indicating the possibility of an inflammatory pathology. Colonoscopy had revealed circumferential non-negotiable distal rectal stricture with ulcerated friable mucosa. Biopsy report and details of treatment were not available.

Laboratory investigations revealed anemia (Hb 8.3 g/dL) while blood counts, biochemistry, serum electrolytes were in normal range. She underwent emergency laparotomy which showed dilated bowel loops up to the sigmoid colon, a collapsed rectum with no evidence of any mass and mild ascites. Sigmoid loop colostomy was performed and per rectal biopsy from the rectal stricture was taken. Peritoneal fluid cytology revealed no malignant cells and biochemistry was suggestive of transudative fluid. Biopsy from rectal stricture revealed chronic non-specific inflammation with trophozoites of Entamoeba histolytica in the wall, suggestive of amoeboma. No evidence of malignancy was seen. Post-operative abdominal CT scan revealed diffuse non-enhancing thickening of rectosigmoid extending over entire colon, up to terminal ileum with maintained mucosal pattern. Part of rectal wall showed circumferential thickening and enhancement (Figure 1). These findings were considered suggestive of an inflammatory pathology and were supported by histopathological findings. Accordingly, oral anti-amoebic drugs were started and patient was discharged.

Two months later the patient was readmitted with intestinal obstruction and managed conservatively. Laboratory investigations were normal apart from raised total counts (12700 /µl), and presence of trophozoites of Giardia lamblia in stool. Ultrasound revealed left sided pelvi-caliectasis and dilated ureter with normal lower abdominal study. CEA was elevated (19.10 ng/mL). Abdominal CT scan revealed findings similar to the previous study, although the pathology seemed to have progressed. Thickening and enhancement of both ureters suggesting urethritis with mild bilateral hydronephrosis was also seen (Figure 2). Biopsy from colonic wall (colostomy site) showed features of amoebic colitis without any evidence of dysplasia or malignancy.

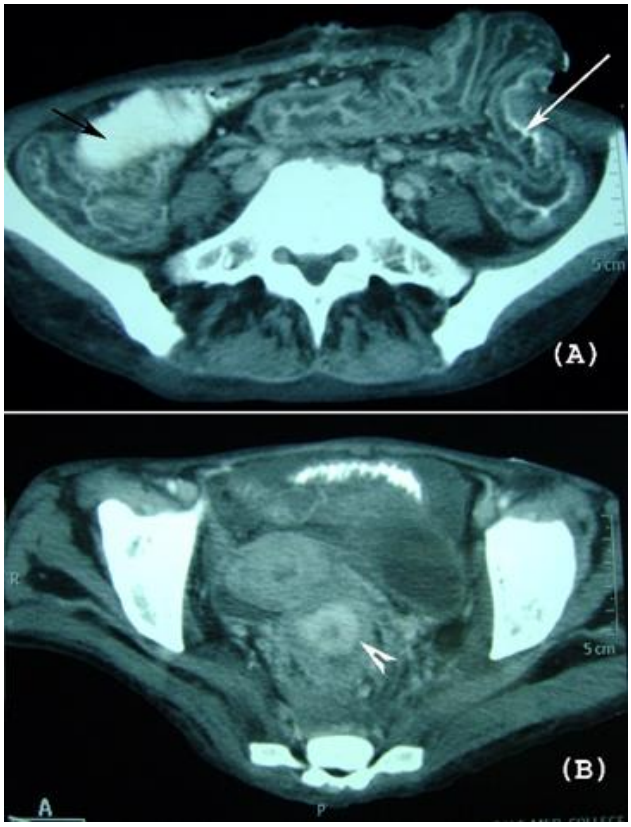

Figure 1 – A) Abdominal CT film after laparotomy and creation of colostomy showing thickened colonic wall with preservation of various layers of the colon (white arrow). Note the relatively normal looking distal ileum (black arrow). (B) Lower cuts in the same CT study showing strictured portion of the rectum (white arrowhead)

Since there was clinical progression of rectal stricture along with apparent infiltration of skin surrounding the colostomy (Figure 3), a third time biopsy from rectal stricture was taken. This revealed presence of signet ring cell adenocarcinoma of rectum (Figure 4). The patient was referred for neo-adjuvant/ primary chemotherapy, after which she was lost to follow up.

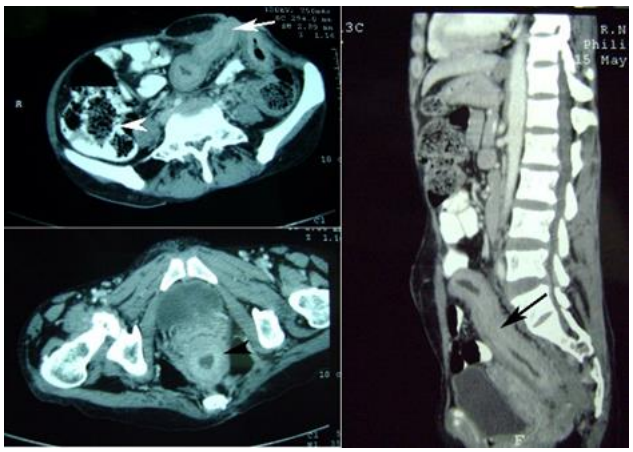

Figure 2 – Abdominal CT performed after 3 months, after having received adequate anti-amoebic treatment. (A) the colonic lesions have considerably subsided, especially in the proximal portions (white arrowhead). The changes are, however, persisting in the distal portion near the colostomy (white arrow). The proximal portion of colon is dilated and loaded with feces indicating colonic obstruction despite the colostomy; (B) strictured portion of the rectum (black arrowhead). (C) Coronal view showing the long segment thickening of distal colon and rectum persisting even after anti-amoebic therapy (black arrow)

Discussions

The case presented herein presented many unique and challenging problems. She presented with rectal stricture leading to acute intestinal obstruction which is an uncommon manifestation of invasive amebiasis. There was invasive amebiasis involving whole of the length of colorectum which is extremely rare. Although malignancy was suspected, repeated biopsies, except the last one, failed to confirm it. The only clue to there being something amiss was the incomplete response to anti-amoebic therapy which we consider as a red flag sign of extreme importance in such a scenario. Diffuse invasive colorectal amebiasis is rare and the present case is probably the first one showing involvement of whole of the colorectum. Due to its rarity it is a challenge to the clinician as well as the radiologist to correctly diagnose such a case.

Localized invasive amebiasis i.e., amoeboma, is uncommon, can be single or multiple and might present as exophytic masses projecting into intestinal lumen, deep penetrating ulcers or strictures [3]. Difficulty in differentiating an amoeboma from carcinoma has long been recognized [3]. It often has an endoscopically similar appearance. Moreover, biopsy is many times inconclusive in neoplasia and stool examination/ cultures/ tissue stains in invasive amebiasis may not show trophozoites. The patient presented herein is an example of this diagnostic difficulty.

Amoebic colorectal stricture is quite uncommon and if it co-exists with other manifestations of colorectal amebiasis, it is of extreme importance to establish whether the stricture is due to amebiasis itself, or is caused by another pathology, most importantly, malignancy.

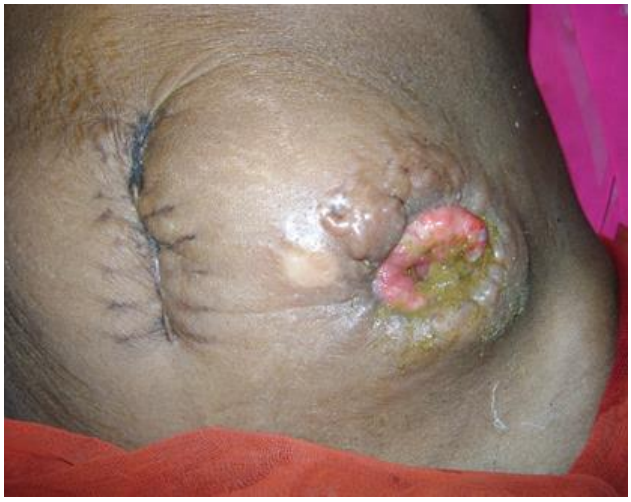

Figure 3 – Clinical photograph of colostomy site showing the thickened stoma along with infiltration of the surrounding skin

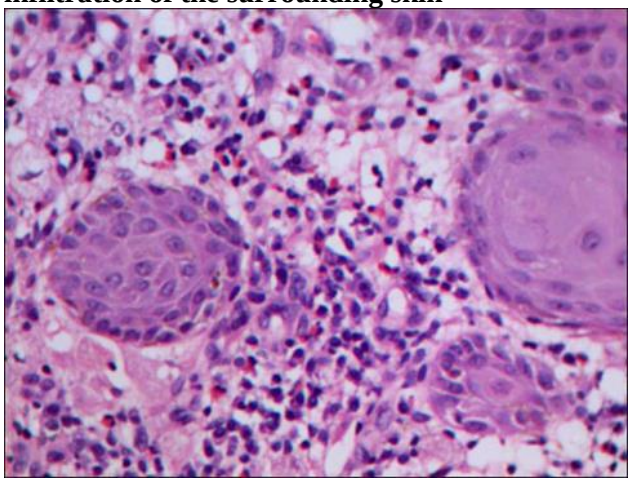

Figure 4 – Photomicrograph showing presence of signet cell adenocarcinoma

The most important radiologic signs described to distinguish between invasive amebiasis and malignancy are multiplicity of lesions, lack of shelving deformity, longer length of the lesion, concentricity, and tapered ends etc. However,

these signs are not completely reliable. Abdominal CT scan findings like diffuse non-specific thickening without other abdominal pathology [3] and conservation of layered structure of colon with marked thickening of submucosal layer of the colon [4,5] have been considered useful in diagnosing invasive amebic colitis. However, confounding findings like apple core appearance has also been reported in invasive amebiasis [6]. Thus, overall, there are no pathognomonic findings establishing a diagnosis of invasive amebiasis [3]. The CT scan findings in the patient presented here are illustrative of radiologic findings in invasive colorectal amebiasis.

Other methods of diagnosing amebic colitis or proctitis are stool examination, presence of trophozoites in biopsy specimen and amebic serology [3]. However, these tests suffer from insufficient sensitivity and do not conclusively rule out presence of co-existent malignancy.

A therapeutic trial with anti-amoebic therapy is commonly used for making this distinction since colorectal amebiasis responds rapidly to it [3,7,8]. When the medical treatment of amebiasis is satisfactory, total regression, including that of colonic amebic stricture [9], is the rule. Although stenosis or stricture due to residual fibrosis has been reported, it is very uncommon [10]. After completion of medical management, follow up sigmoidoscopy or colonoscopy is considered essential to ensure complete resolution [3].

In the present case, the first two biopsy reports had established the presence of amebic colitis, but there was incomplete response to anti-amoebic therapy evidenced by progression in distal colon although there was regression in distal ileum and proximal colon. This alerted us to the possibility of another pathology, leading to another biopsy being performed, which finally established the crucial missing piece of diagnosis.

In our patient, residence in endemic area, absence of family history of colorectal cancer, leukocytosis, involvement of long segment of intestine, diffuse thickening of the bowel wall and response to anti-amoebic treatment, albeit partial, supported the primary diagnosis of amebic colitis. However, the primary presentation with intestinal obstruction, and incomplete resolution with anti-amoebic therapy suggested another pathology.

Such a case presents a dilemma in management as well. The presence of amebic pathology, by confounding the diagnosis can lead to serious delay in instituting treatment for the malignant lesion. In addition, it is difficult to operate or subject the patient to chemoradiation without definitive diagnosis of malignancy. Fortunately, these patients are reported to respond rapidly to anti-amoebic therapy [2]. However, if the response is not rapid enough, this too shall lead to delay in treating the malignancy. Finally, in case the colitis fails to respond completely, alteration in management plan or operative procedure itself might become necessary.

It is highly risky to operate a patient with untreated invasive amebiasis because of high attendant morbidity and mortality (up to 50%) [6,8,11]. Surgical intervention is only indicated in rare instances of acute necrotizing colitis with perforation [3]. Therefore, it is very important to recognize invasive amebiasis, especially in areas endemic for E. histolytica. Even in countries traditionally non-endemic for amebiasis its incidence has increased to an extent where it should be kept as a differential diagnosis for colorectal diseases, notably malignancy.

Conclusion

We propose that in cases where amebic pathology is suspected, but either the presentation or clinical features suggest possibility of malignancy, diagnostic tests for amebiasis should be performed pari-passu with investigations to rule out malignancy. Even if amebiasis is confirmed and malignancy ruled out, a therapeutic trial of anti-amoebic medication should be given. If the response is not adequate, proper biopsy, repeated if required, should be performed. This should be especially so if the involvement is of localized variety, namely a mass or a stricture.

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