International Journal of Cancer Therapy and Oncology
www.ijcto.org

Xanthogranulomatous colitis masquerading as carcinoma of colon

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Received January 26, 2015; Revised January 30, 2015; Accepted January 31, 2015; Published Online February 20, 2015

Case report

Abstract

Xanthogranulomatous inflammation (XGI) is an uncommon pathological diagnosis involving various organ systems, the most common being the gall bladder and kidney. It can masquerade as a malignant mass thus, requiring a clinical suspicion for accurate and timely diagnosis. A 65-year-old woman presented with acute onset of obstipation and vomiting suggesting acute obstruction. Contrast enhanced computed tomography of abdomen revealed a solid irregular mass in the ascending colon with large necrotic areas and surrounding enlarged nodes suggestive of malignancy arising from right colon. Right hemi-colectomy was performed. Histopathology of the surgical specimen showed florid inflammatory infiltrate with collection of histiocytes, lymphocytes and polymorphs. Further immunohistochemistry was conducted, and CD68 and CD45 were found to be positive and pan-cytokeratin was negative. A clinico-pathological diagnosis was thus established to be xanthogranulomatous colitis.

Keywords: Xanthogranulomatous Inflammation; Colitis; Immunohistochemistry; Colon Cancer

Introduction

Xanthogranulomatous inflammation (XGI) is an uncommon pathological diagnosis involving various organ systems, the most common being the gall bladder and kidney.[1, 2] XGI of the gastrointestinal tract (GI-XGI) has rarely been reported.[3] The etio-pathogenesis of GI-XGI is not well understood, the normal structures in the involved organ may undergo destruction and effacement, and appear as a locally invasive malignant lesion.[4] This clinio-pathological finding is not considered to be a premalignant condition. In this report, we present a rare case of XGI of ascending colon which was initially interpreted as a case of carcinoma of right colon.

Case presentation

A 65-year-old woman presented with acute onset of obstipation and vomiting suggesting acute obstruction. The patient had history of constipation along with on-and-off pain abdomen for 2 months. There was no history of hematemesis or melena. No addiction history was present. On examination, the vitals were normal; however, abdomen was tender particularly in the right lower quadrant. No other significant findings were present. Hematological tests suggested microcytic, hypochromic anemia with haemoglobin 9.8 g/dL. Biochemical tests were unremarkable except mildly elevated aspartate aminotransferase (53 U/L) and alanine aminotransferase (64U/L). Contrast enhanced computed tomography (CE-CT) of abdomen and pelvis revealed a solid irregular mass in the ascending colon, measuring 84 mm × 87 mm × 56 mm, with large necrotic areas and surrounding enlarged nodes suggestive of malignancy arising from right colon (**Figure 1**). Carcino-embryonic antigen value was within normal limits (3 IU/ml). Median laparotomy was performed due to worsening of pain despite conservative management. Intraoperative findings included a large mass, involving the ileocecal valve and fixed to the nearby structures. Right hemi-colectomy was performed, the patient was stable post-operatively and the symptoms were relieved. Histopathology of the surgical specimen showed florid inflammatory infiltrate with collection of histiocytes, lymphocytes and polymorphs (**Figure 2A**). Stains for Acid fast bacillus and fungus were carried out with adequate controls and were negative. Further immunohistochemistry was conducted, and CD68 (**Figure 2B**) and CD45 (**Figure 3A**) were found to be positive and pan-cytokeratin was negative (**Figure 3B**). A clinico-pathological diagnosis was thus established to be xanthogranulomatous colitis. The patient is on follow-up and symptom free from six months after surgery.

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***Cite this article as**: Kapoor A, Soni D, Paramanandhan M, Kini L, Beniwal S, Kumar H. Xanthogranulomatous Colitis masquerading as carcinoma of colon. Int J Cancer Ther Oncol 2015; 3(2):03025. **DOI:** 10.14319/ijcto.0302.5*

ISSN 2330-4049

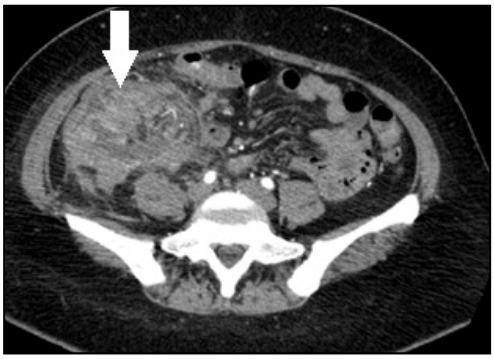

FIG. 1: Contrast enhanced computed tomography of abdomen showing a large mass in the right side.

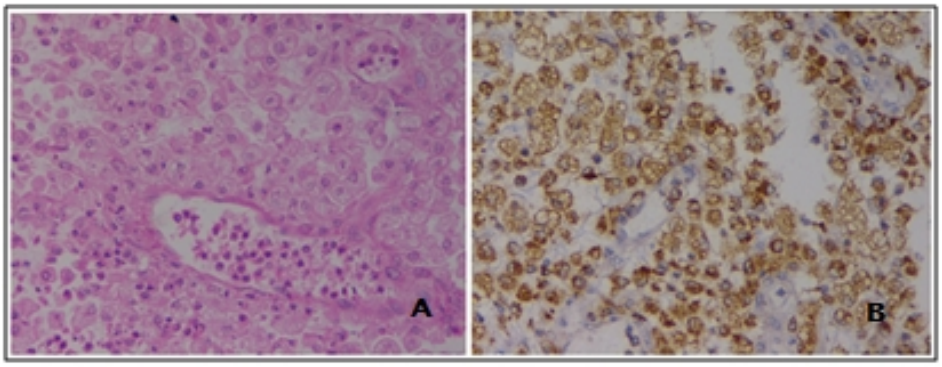

FIG. 2: Hematoxylin and eosin staining of the surgical specimen showed florid inflammatory infiltrate with collection of histiocytes, lymphocytes and polymorphs (Figure 2A, X 400) and positive staining for CD 68 (Figure 2B, X 400).

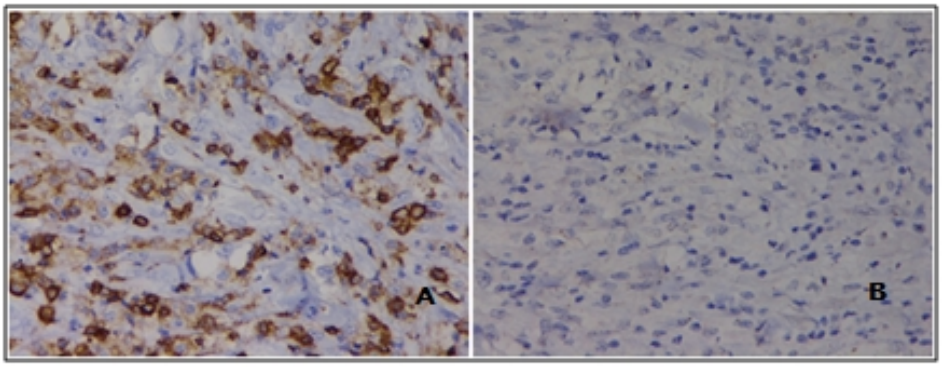

FIG. 3: Immunohistochemistry study showing positive staining for CD 45 (Figure 3A, X400) and negative staining for pan-cytokeratin (Figure 3B, X 160).

Discussion

Xanthogranlumatous colitis is a rare presentation of XGI.[5] The most common location include gall bladder and kidney. The other sites include the pancreas, appendix, eyes, female genital tracts, and urachus. There is no clear age or sex preponderance, there have been few cases reported in paediatric age as well.[6] The reported case under discussion is an elderly female in her 7th decade. XGI disease has a variable presentation; both localized and diffuse responses can be seen. The more severe form of disease is a diffuse inflammation, which clinically presents as a solid mass. This occurs due to the complete blockage of the original parenchyma of the tissue. XGI can also have a cutaneous involvement which can be primary or secondary.[7] The secondary disease is a manifestation of a very severe underlying disease process. Clinically, patients present as fistulas on the skin with a track extending to the involve organ. These tracks can become infected and foul smelling discharge can also be seen. In the present case study, the patient presented with features suggestive of acute obstruction viz. obstipation and vomiting.

The etio-pathogenesis of the disease is not well understood; the probable hypothesis could be organ obstruction, suppurative infections, defective lipid transport, immunological and haemorrhage which may trigger tissue damage within

the involved organs, a microscopic reaction of the disease process sets in. The infective etiology of this condition can be substantiated by case study of Luc et al, as seen following chronic infections with mycoplasma hominis.[8] These triggers, either alone or together can produce an extensive long standing chronic inflammation of the organ involved leading to xanthogranulomatous transformation. In xanthogranulomatous appendicitis, a similar obstructive etio-pathology can explain the chronic inflammatory changes in the lumen of the vermiform appendix following obstruction of the lumen with a faecolith. The other etiologies hypothesised include sub-mucosal dissection, mucosal resection or pre-operative endoscopic biopsy. The probable mechanism is that endoscopic procedure may cause damage to the mucosa, leading to bacterial invasion at the site of damage. The gastric acid and other secretions along with the bacterial infection can set a complex irreversible pathological process causing XGI. In our case, no such risk factor was present.

On gross examination, the lesion appears as a bright yellow or solid golden yellow with ill-defined white areas of fibrosis as in our case. Histology shows characteristic adequate numbers of foamy histiocytes and multinucleated giant cells with inflammatory cells and a fibrous reaction. Few areas show necrosis with eosinophils and a fibrous reaction. Immunohistochemistry reveals positivity for CD68, but negative for CD 117, S-100, and cytokeratin as in our case. Periodic acid schiff, Gomori's methenamine silver and acid-fast stains are negative. Nontuberculous mycobacterium and mycobacterium tuberculosis-PCR from the representative paraffin embedded tissue are usually negative.[9] These yellow lesions should be differentiated from a pseudo-xanthomatous inflammation or malakoplakia. Michaelis-Gutman bodies are characteristic on microscopy, and stain is positive for Prussian blue and Von-Kossa calcium stains.

Radiological diagnosis of XGI using ultrasound scan and computer tomography imaging is a challenge. A diagnosis of a malignant lesion can be concluded based on endoscopic, ultrasonographic and radiological findings instead of GI-XGI.[9] Diagnostic accuracy can be improved by the use of contrast-based USG and contrast enhanced computer tomography to demonstrate XGI.[10] The characteristic findings on imaging are the areas of low attenuation nodules which corresponds to the xanthomatous changes on histology. On endoscopy, a typical picture of ulcerative mucosa with underlying sub-mucosal lesion is seen. Also, abdominal computed tomography (CT) shows a mass-like lesion in the gastrointestinal wall occasionally.[11] Contrast CT scan in the present case showed a solid irregular mass in the ascending colon, with large necrotic areas and surrounding enlarged nodes suggestive of malignancy arising from right colon. Thus, radiological findings alone may lead to misdiagnosis. **Table 1** describes the reported cases of large bowel XGI.

TABLE 1: Reported cases of large bowel xanthogranulomatous inflammation.

Ref	Age (yr) / sex	Involved organs	Clinical presentation	CT findings	Surgical operation/ pathology
Davis, *et al.*[12]	30/F	Anorectal area	Pain in recto-sacral area with nausea and vomiting, per-rectal examination revealed tender palpable mass.	CT: two masses in the rectal region infiltrating the intramural areas with extension to involve the sacrum.	Posterior approach, removal of the tumors, re-approximation of the rectal wall.
Lo, *et al.*[13]	72/M	Sigmoid colon	Nausea, vomiting, tenesmus, intestinal obstruction with tenderness in LIF.	Barium enema: obstruction secondary to stricture in sigmoid colon.	Excision of the mass including sigmoid colon, proximal jejunum and part of the peritoneum.
Oh, *et al.*[14]	38/F	Sigmoid colon, appendix, salpinx	Fever, abdominal pain, diarrhea with mild RLQ tenderness without any palpable mass.	CT: mass in recto-sigmoid junction with loops of colon and infiltration of mesenteric fat.	Radical resection of the appendix, sigmoid colon and right salpingectomy.
Anadol, *et al.*[15]	57/F	Cecum and appendix	RLQ abdominal pain, pallor, haemotochezia.	CT: Solid mass in the RLQ with ill-defined margins and infiltration of mesenteric fat.	Appendectomy and right colectomy; found to be XGI of the cecum.
Dhawan, *et al.*[5]	60/M	Ascending colon	Abdominal pain, constipation, vomiting	CT: involvement of 6 cm of ascending colon with irregular circumferential thickening and multiple pericolic lymph nodes.	Right colectomy; found to be XGI of the ascending colon with mucosal involvement.
Present case	65/F	Cecum, jejunal loop	Abdominal pain, palpable mass in the RIF, fever and weight loss	CT: solid irregular mass in the asce- nding colon, measuring 84mm × 87 mm × 56mm, with large necrotic areas and surrounding enlarged nodes	Right hemi-colectomy; found to be XGI of the ileocecal valve.

CT = Computed tomography; M = Male; F = Female; NA = Not available; XGI = Xanthogranulomatous inflammation; RLQ = Right lower quadrant; LIF = Left iliac fossa; RIF = Right iliac fossa.

Management includes complete surgical resection, followed by observation. Complete resection negates the need for any further definitive management as this is not a pre-malignant condition. There are no recurrences reported in the primary or other organs following the surgical treatment. Generally prognosis is very good, if there are no co-morbid factors. As the radiological studies pointed towards malignancy, we preferred a radical approach in the treatment; the patient underwent an uneventful hemi-colectomy. The patient is doing well on observation alone post-surgery.

Conclusion

Xanthogranulomatous colitis can masquerade as a malignant mass thus, requiring a clinical suspicion for accurate and timely diagnosis.

Conflict of interest

The authors declare that they have no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

References

1. Kansakar PB, Rodrigues G, Khan SA. Xanthogranulomatous cholecystitis: a clinicopathological study from a tertiary care health institution. *Kathmandu Univ Med J (KUMJ)* 2008; **6**:472-5.
2. Siddappa S, Ramprasad K, Muddegowda MK. Xanthogranulomatous pyelonephritis: a retrospective review of 16 cases. *Korean J Urol* 2011; **52**:421-4.
3. Jung SH, Kim JH. Cecal cancer with xanthogranulomatous inflammation. *J Korean Surg Soc* 2008; **74**: 392-5.
4. Goodman ZD, Ishak KG. Xanthogranulomatous cholecystitis. *Am J Surg Pathol* 1981; **5**:653-9.
5. Dhawan S, Jain D, Kalhan SK. Xanthogranulomatous inflammation of ascending colon with mucosal involvement: report of a first case. *J Crohns Colitis* 2011; **5**:245-8.
6. Bouali O, Faure A, Chaumoitre K, *et al.* Diffuse xanthogranulomatous pyelonephritis in infant. *Prog Urol* 2011; **21**:495-9.
7. Rogers S, Slater DN, Anderson JA, Parsons MA. Cutaneous xanthogranulomatous inflammation: a potential indicator of internal disease. *Br J Dermatol* 1992; **126**:290-3.
8. Biedermann L, Schaer DJ, Montani M, *et al.* Extensive chronic xanthogranulomatous intra-abdominal inflammation due to Mycoplasma hominis mimicking a malignancy: a case report. *J Med Case Reports* 2009; **3**:9211.
9. Hwang SW, Lee SJ, Park PE, *et al.* Xanthogranulomatous inflammatory reaction associated with endoscopic mucosal resections of gastric and colonic cancer. *Basic and Applied Pathology* 2011; **4**:125-9.
10. Ueda J, Yoshida H, Arima Y, *et al.* A case of xanthogranulomatous cholecystitis preoperatively diagnosed with contrast-enhanced ultrasonography. *J Nippon Med Sch* 2011; **78**:194-8.
11. Kubosawa H, Yano K, Oda K, *et al.* Xanthogranulomatous gastritis with pseudosarcomatous changes. *Pathol Int* 2007; **57**: 291-5.
12. Davis M, Whitley ME, Haque AK, *et al.* Xanthogranulomatous abscess of a mullerian duct remnant. A rare lesion of the rectum and anus. *Dis Colon Rectum* 1986; **29**:755-9.
13. Lo CY, Lorentz TG, Poon CS. Xanthogranulomatous inflammation of the sigmoid colon: a case report. *Aust N Z J Surg* 1996; **66**:643-4.
14. Oh YH, Seong SS, Jang KS, *et al.* Xanthogranulomatous inflammation presenting as a submucosal mass of the sigmoid colon. *Pathol Int* 2005; **55**:440-4.
15. Anadol AZ, Gonul II, Tezel E. Xanthogranulomatous inflammation of the colon: a rare cause of cecal mass with bleeding. *South Med J* 2009; **102**:196-9.

ISSN 2330-4049