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Ascending Colon Leiomyoma With Osseous Metaplasia Resembling Uterine Fibroid: A Case Report

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Case Report

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Abstract

Background

Colonic leiomyoma with osseous metaplasia is an uncommon finding in colonic leiomyomas with unclear etiology. The rarity of the lesion, its radiologic and histologic mimicry of malignancy (notably GIST and leiomyosarcoma), and the presence of heterotopic bone formation complicate preoperative diagnosis. Treatment decisions are often more aggressive than necessary due to diagnostic uncertainty.

Case presentation

We presented a 32years old African female, with progressive abdominal distension for 4 months, associated with colicky abdominal pain. On examination the abdomen was asymmetrically distended with irregular palpable huge mass more localized on the right lower quadrant which was firm in consistent, and non-tender. Fluid thrill was negative. Examination other systems were unremarkable. After surgical resection of the huge mass arising from ascending colon muscularis layer, histomorphological and immunohistochemically it was consistent with leiomyoma with osseous metaplasia. Intraoperative findings were a huge tumor arising from the colonic wall and Right hemicolectomy was done followed by end to ileo-transverse anastomosis. Outcome was generally excellent after complete excision and the patient had two surgical clinic follow-up visits.

Conclusion

Colonic leiomyoma with osseous metaplasia is clinically significant despite its benign nature because it can closely mimic malignant colonic tumors on imaging, endoscopy, and gross examination due to its firmness and calcified/ossified components. Awareness of this entity is essential for pathologists and clinicians to ensure accurate immunohistochemical diagnosis, avoid misclassification as GIST or leiomyosarcoma, and prevent unnecessary aggressive surgery

INTRODUCTION

Leiomyoma is a benign smooth muscle spindle cell proliferations which primarily occur in female genital tract. Rarely leiomyoma occur in the colon arising from muscularis mucosae, muscularis propria or vascular smooth muscles of the gastrointestinal tract (GIT) especially at esophagus and stomach(1), followed by small intestine and rectosigmoid region(1). Colonic leiomyoma are common in men than women with male to female ratio of 2.4:1 and median age of diagnosis is 62years(1) and accounts for 3% of GIT leiomyoma's(1). Descending and sigmoid colon are the most frequently involved region of colon(2).

Most patients are incidentally diagnosed during imaging tests or endoscopic evaluation for other conditions, however small percentage of patients may present with symptoms like constipation, abdominal pain, intestinal obstruction, occult bleeding, intestinal perforation or palpable intraabdominal mass(2–4). Other clinical presentations depends on size and mass effect of the lesion particularly for those masses which are more than 15cm size(2). The risk factors for colonic leiomyoma are unknown but some literature reported to be associated with inflammatory bowel diseases (IBD)(2). Pathogenesis of osseous metaplasia remains unclear, but hypothesized mechanisms include: Chronic ischemia or degeneration, Local inflammation inducing mesenchymal stem cell differentiation, Bone morphogenetic protein (BMP) expression within smooth-muscle tumors(5, 6)

Colonic leiomyoma is diagnosed by colonoscopy, imaging methods like computed tomography (CT) with colonography or magnetic resonance imaging (MRI. Histomorphological confirmation under the light microscope is a gold standard of diagnosis and is helpful in ruling out of polyps or gastrointestinal stromal tumours (GIST) particularly, with complement of immunohistochemical tests in some cases(4, 7). Endoscopically they may resemble colonic adenomas, or solitary intraluminal or intramural polyps (semi-pedunculated, pedunculated or sessile polyps(7, 8). They are removed by either endoscopic resection if they are intraluminal or segmental colectomy especially when they are larger lesions or arising intramural. Colonic leiomyoma are associated with very good prognosis without risk of malignant transformation or recurrence after complete surgical removal(9). Follow up after surgery is paramount if histomorphological cytological atypia is observed. We presented a case of histologically confirmed colonic leiomyoma with osseous metaplasia in 32 yrs old female because Leiomyoma with osseous metaplasia in the colon poses disproportionate diagnostic and management challenges despite its benign biology. The rarity of the lesion, its radiologic and histologic mimicry of malignancy (notably GIST and leiomyosarcoma), and the presence of heterotopic bone formation complicate preoperative diagnosis. Treatment decisions are often more aggressive than necessary due to diagnostic uncertainty. Outcomes are generally excellent after complete excision, but long-term follow-up data are scarce, limiting evidence-based prognostication(10, 11)

CASE REPORT

A 32 years African female presented at Bukoba Regional Referral Hospital surgical department with a complaint of abdominal distension of gradual onset for 4 months, progressively increasing in severity with time, which was associated with localized colicky abdominal pain more on the right lower quadrant. She denied history of nausea, vomiting, constipation, diarrhea or passing blood-stained stools. Also, she denied any history of surgery, family history of the same illness or inherited diseases. Other systems were unremarkable. On physical examination the abdomen was asymmetrically distended, moving with respiration, with palpable irregular huge mass, originating from the pelvic region occupying the entire right lower quadrant, it was mobile, non-tender, firm to hard. Fluid thrill was negative. Laboratory tests which were done which includes CA-125 which was within normal limits, urine pregnancy test (UPT) was negative, and complete blood count (CBC) in which all parameters were within normal limits except for hemoglobin level which was 8.0g/dl. Abdominal pelvic CT scan revealed huge intraabdominal tumour

measuring 27.0x21x17 cm with areas of cystic degeneration. And pre operative diagnosis was possible leiomyoma arising from uterine serosa. Intraoperative findings were a huge tumor arising from the colonic wall and Right hemicolectomy was done followed by end to ileo-transverse anastomosis. And the post operative diagnosis was leiomyoma arising from the colon. The specimen was submitted to histopathology and morbid anatomy Bugando medical center (BMC) zonal Referral Hospital. Gross examination found a right hemicolectomy specimen with capsulated irregular mass firm to hard of 28x22x18 cm originating from the wall of the ascending colon. The mass was 2 cm from ascending colon surgical resection margin and 5cm from ileal surgical resection margin. The specimen was attached with 6 cm long appendix which was not involved by the mass. On cut section the lumen of ascending colon was patent with normal mucosal layer. Ileal and appendix lumens were macroscopically normal. On further cut section the mass had solid areas, which were firm to hard and homogenous white in color, accounting for approximately 70% of the specimen and areas of multiloculated cystic degenerations containing brownish gelatinous materials (see Fig. 1) accounting for the remaining percentage. Histologically representative sections showed benign spindle cell lesion forming fascicles of interlacing bundles, with cigar shaped nuclei and blunt ends. Elsewhere there were areas of matured bone formation containing osteoblastic and osteoclastic cells associated with multiple osteoclastic giant cells. These features were consistent with leiomyoma (symplastic type) with osseous metaplasia (see Figs. 2 and 3). IHC smooth muscle actin (SMA) and CD117 were done, in which SMA was positive confirming smooth muscle differentiation of the tumour cells and CD117 was negative rulling out GIST (See Fig. 4). Post hospital discharges the patient had two surgical clinic follow-up visits, of which the last was three months after surgery, where she underwent some investigations including abdominal pelvis ultrasound which revealed normal findings

DISCUSSION

Colonic leiomyoma account for 3% of GIT leiomyoma's of which in the literature none was reported to present with osseous metaplasia unlike their counterpart in female genital tract which may show osseous(12–14) or chondroid differentiation(14). Other degenerative changes in leiomyoma like hyaline change, cystic change, dystrophic calcifications, and infiltration of inflammatory cells or osseous metaplasia are common in uterine and extra uterine lesions(12, 13, 15, 16). In our case the lesion was arising from muscularis mucosae of ascending colon and histologically extensive areas of osseous metaplasia without cytological atypia were appreciated. The exact cause of osseous metaplasia in leiomyoma is unknown although literature suggest that is due to reprogramming of undifferentiated mesenchymal cells under the influence of cytokines, growth factors and extracellular matrix component within the cellular environment(17). The top on the list of differential diagnoses for this case were leiomyosarcoma and GIST(18). Histomorphogically the absence of frequent mitosis, coagulative necrosis, pleomorphism of spindle cells or significant cytological atypia ruled out leiomyosarcoma which is the malignant counter part of leiomyoma(2). Similarly, histomorphological absence of intersecting fascicles of plump spindled cells or rounded epithelioid cells with pseudo compartmental organization and skeinoid fibers ruled out GIST.

Immunohistochemically colonic leiomyoma expresses smooth muscle actin (SMA), desmin and H-caldesmon, however they are negative for CD117 (c-kit), CD34 and S-100 protein. These immunohistochemistry tests are important in distinguishing leiomyoma from other colonic smooth muscle tumour(9). For our case SMA was positive confirming leiomyoma and CD117 was negative rulling out GIST.

The purpose of this case report was to highlight and document an extremely rare histopathological feature which have been rarely reported in colonic leiomyoma to increase awareness among pathologists, surgeons and gastroenterologist.

CONCLUSION

Colonic leiomyoma with osseous metaplasia is clinically significant despite its benign nature because it can closely mimic malignant colonic tumors on imaging, endoscopy, and gross examination due to its firmness and calcified/ossified components. This frequently leads to diagnostic uncertainty and overtreatment, including segmental colectomy rather than limited excision. Awareness of this entity is essential for pathologists and clinicians to ensure accurate immunohistochemical diagnosis, avoid misclassification as GIST or leiomyosarcoma, and prevent unnecessary aggressive surgery. Once correctly diagnosed and completely excised, the lesion carries an excellent prognosis with negligible risk of recurrence or malignant transformation, though evidence-based surveillance strategies are lacking due to rarity

Declarations

Competing interests

The authors have no competing interest to declare that are relevant to the content of this article

Ethics approval of the institutional IRB

Not applicable at our institution

Consent to participate

Written informed consent was obtained from the patient to participate in this case report. A copy of the written consent is available for review by the Editor-in-chief of this journal

Consent to publish

Written informed consent was obtained from the patients next of kin for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-chief of this journal

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Authors' contributions

Conceptualization: AHC and SM; writing—original draft preparation: MN and EM; investigations especially radiology: AHC; writing and editing: PR; supervision. All authors reviewed and approved the final manuscript

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Figures

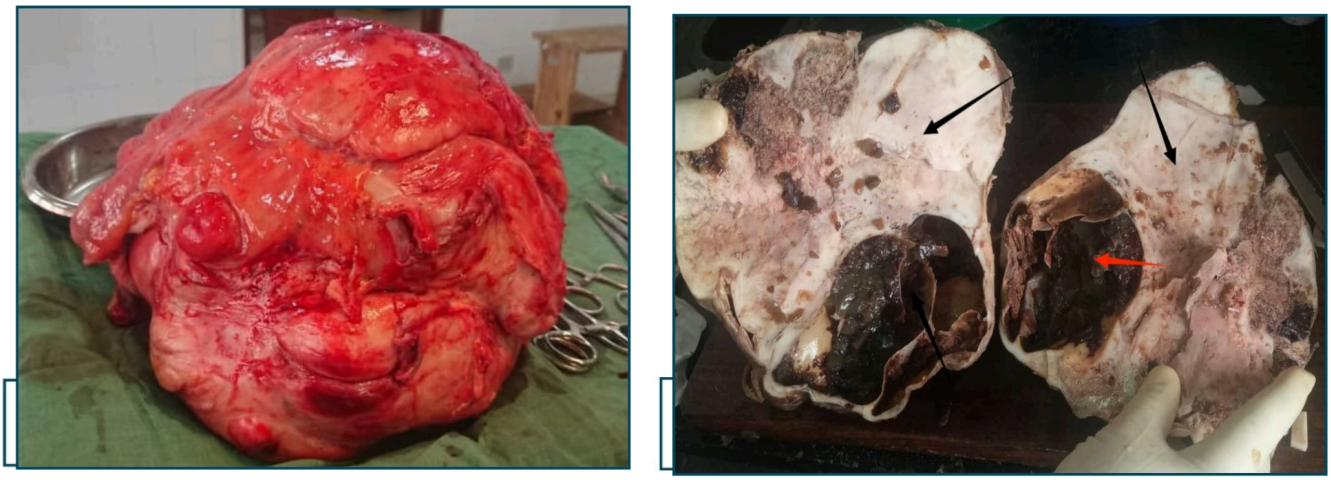
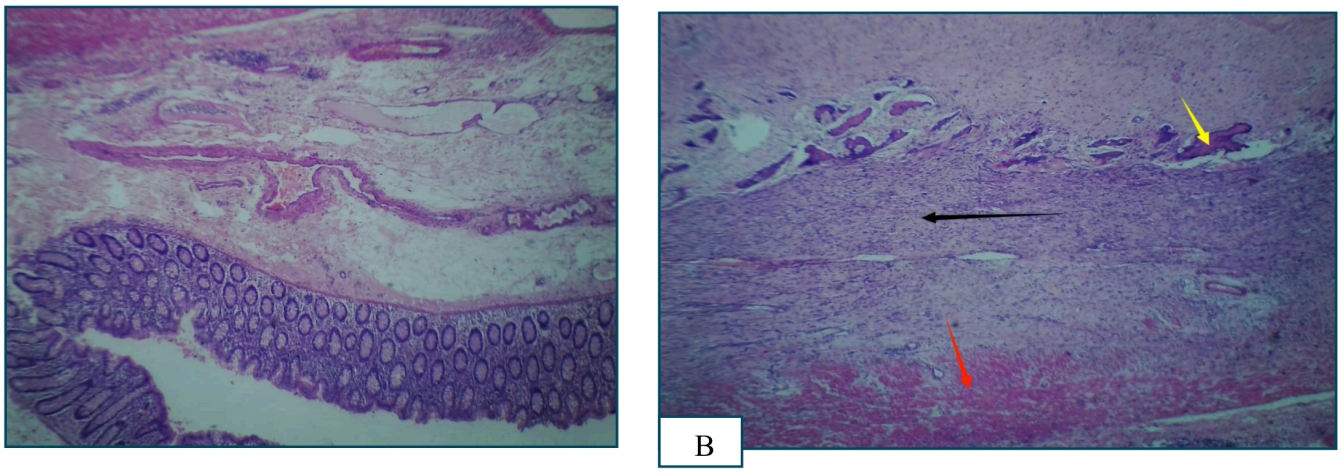


Figure 1

A; unfixed right hemicolectomy specimen immediately after surgery. B; Formalin fixed specimen with solid areas of about 70% (black arrows) and about 30% of cystic degeneration with cystic lumen containing brown gelatinous mucoid material (red arrows).



B

Figure 2

A: H &E section (40X) showing normal lining epithelium with intestinal glands without atypia and submucosa layer. B: Spindle cell proliferation (black arrow) arising from the muscularis propria of the colonic wall (red arrow) with mature bone (yellow arrow) (H&E 40X)

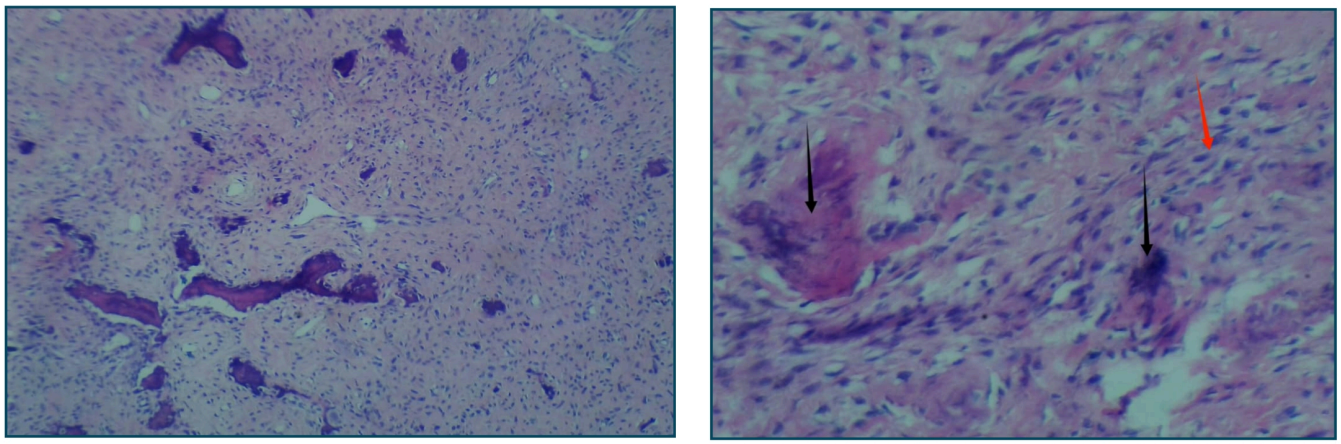


Figure 3

A: H&E section (100X) showing fascicle of interlacing bundles with cigar shaped nuclei and blunt ends and additional extensive areas of mature bone trabecular both without atypia. B: H&E section (200X) showing fascicle of interlacing bundles with cigar shaped nuclei (red arrow) and blunt ends and additional extensive areas of ossification (black arrows).

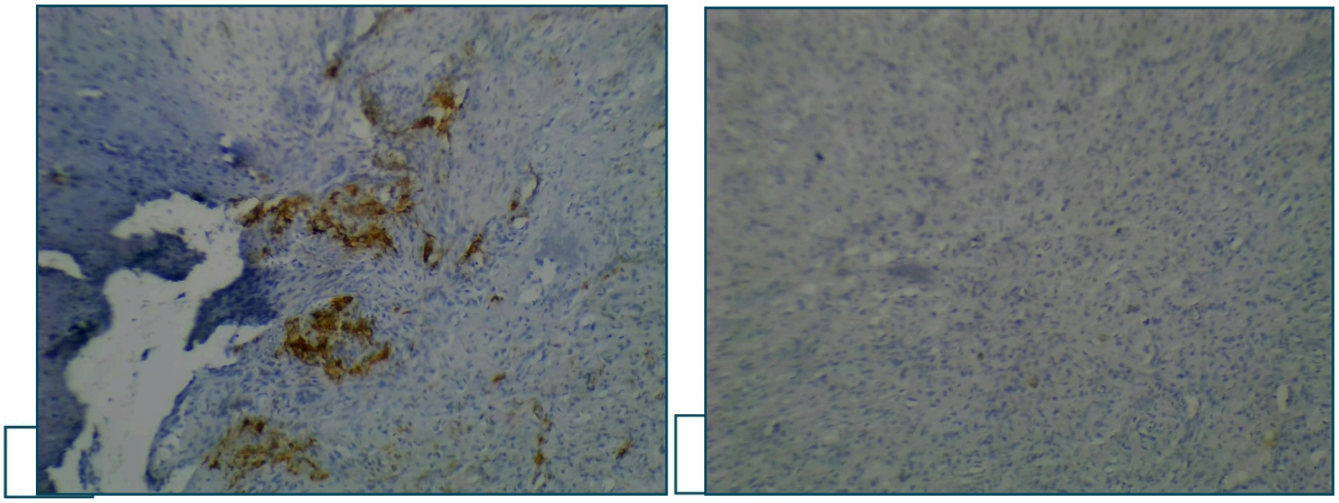


Figure 4

A; Immunohistochemical staining of SMA showing focal and strong cytoplasmic staining of tumor cells. B; Immunohistochemical staining of CD117 showing negative staining of tumor cells