J Gastrointest Canc (2007) 38:160–163
DOI 10.1007/s12029-008-9031-1

Primary Colonic Liposarcoma Causing Colo-colic Intususception: A Case Report and Review of Literature

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Published online: 30 October 2008
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Abstract
Introduction Liposarcomas are most common soft tissue sarcomas usually seen in deep soft tissues of extremities and retroperitoneum. Although secondary involvement of gastrointestinal system can occur in cases of retroperitoneal tumours, liposarcomas primarily involving the gastrointestinal tract are extremely uncommon. Intusussception refers to telescoping of contiguous segments of intestine causing obstruction. Colo-colic intususception is rare in adults and usually associated with a malignant etiology, usually epithelial malignancies (adenocarcinomas).
Discussion Colonic liposarcomas leading to colo-colic intussusception are extremely rare, with only six cases reported in the literature.
Conclusion In this report, we describe this rare occurrence in a 66-year-old lady and highlight the diagnostic difficulties such cases can pose along with a review of relevant literature.

Keywords liposarcoma · intususception · gastrointestinal · colonic

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Introduction

Liposarcomas are most common soft tissue sarcomas commonly seen in the retroperitoneum and deep soft tissues of trunk and extremities. Occurrence of these tumours in the gastrointestinal system is uncommon and usually involves the small intestine. Primary colonic liposarcomas are extremely rare, and only six previous cases have been described in the English literature [1–6]. Owing to their rarity, the mode of presentation and true biologic behaviour of these lesions is not well known.

Intusussception refers to telescoping of contiguous segments of intestine into one another leading to bowel obstruction. Whilst this is relatively common in children, it accounts for only 1–3% cases of adult bowel obstruction [7]. In addition, whilst majority of childhood intussusceptions are idiopathic, about 90% of adult bowel obstructions have a demonstrable etiology.

Colonic intussusception (colo-colic, colorectal or sigmoidorectal) in adults is rare and comprises less than one third of all such cases. A malignant aetiology is seen in about 75% of colonic intussusceptions in contrast to small bowel or ileocolic obstructions which have a malignant etiology in only about 25% of cases [8].

Case Report

A 66-year-old lady presented with acute onset pain in abdomen of 2days duration. On closer questioning, she gave history of altered bowel habits and history of passage of blood and mucus in stools for 6 months. There was no history of anorexia or significant weight loss. Systemic examination revealed a 6×3-cm non-tender, firm lump in left iliac fossa. Routine laboratory investigations were

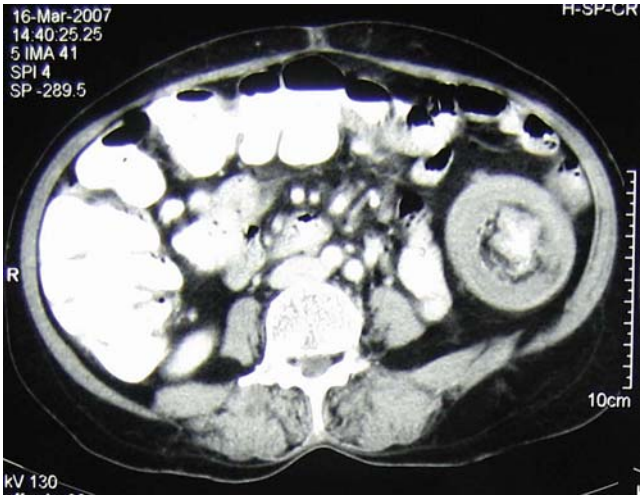

Fig. 1 Contrast-enhanced CT showing colo-colic intususception and an intraluminal mass showing heterogenous contrast enhancement

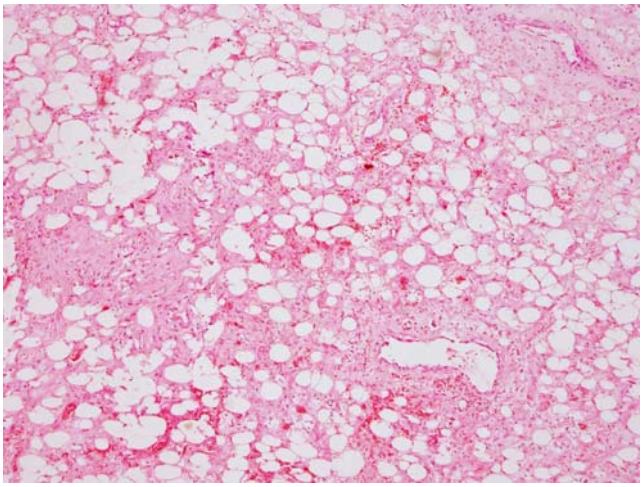

Fig. 3 Photomicrograph showing a tumour composed predominantly of mature adipose tissue (haematoxylin & eosin ×100)

within normal limits except for presence of anaemia (haemoglobin 7.0 gm/dl).

Ultrasound examination of abdomen showed features suggestive of a colo-colic intususception. Contrast-enhanced tomography (CECT) confirmed this finding and revealed an intraluminal mass lesion in the distal descending colon which exhibited heterogenous contrast enhancement. There was no associated regional lymphadenopathy (Fig. 1).

With this clinico-radiological picture, an exploratory laparotomy was performed. Intraoperatively, there was colo-colic intususception with descending colon telescoping into the sigmoid colon. An intraluminal pedunculated lesion measuring about 4×3 cm was seen in the descending colon (Fig. 2). The intussusception was reduced and a left hemicolectomy was performed. Postoperative course was uneventful.

A left hemicolectomy specimen measuring 20 cm was received. Cut section revealed an intraluminal polypoidal mass measuring 4.5×3×2 cm. Adjacent colonic mucosa was grossly unremarkable. The cut section of the tumour was pale yellow with fine fibrous septae traversing the lesion.

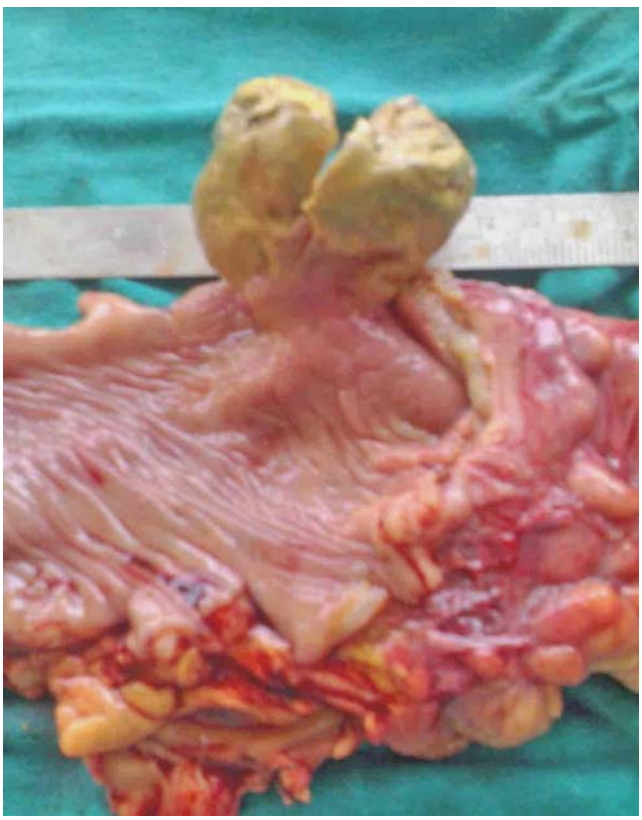

Fig. 2 Gross photograph showing a pedunculated intraluminal mass in the descending colon

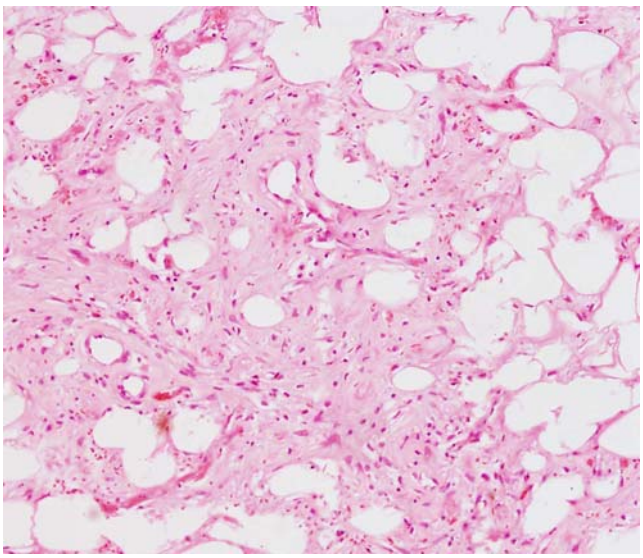

Fig. 4 Photomicrograph showing presence of spindle cells exhibiting mild nuclear pleomorphism. (hematoxylin & eosin ×200)

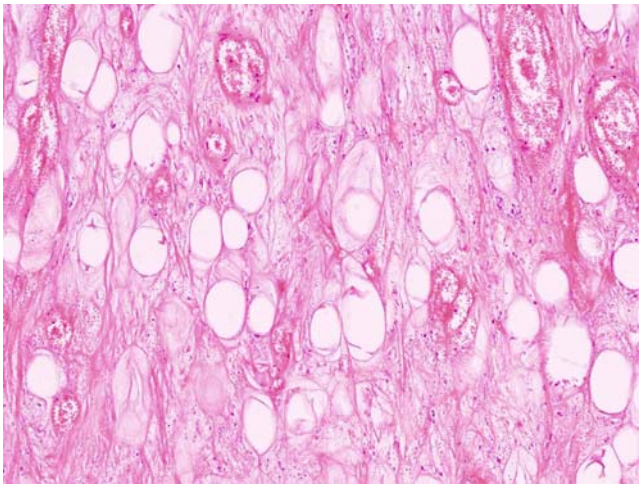

Fig. 5 Photomicrograph showing an area of coagulative necrosis. (hematoxylin & eosin ×200)

Microscopic examination showed a submucosal lesion comprising predominantly of mature fat-like areas (Fig. 3). However, at places, a variable number of spindle cells showing mild degree of nuclear pleomorphism were noticed (Fig. 4). Mitotic figures were rare. No lipoblasts were identified. In addition, there were focal areas of coagulative necrosis (Fig. 5). Overlying mucosa showed areas of ulceration. Immunohistochemically, the tumour cells were positive for S-100 and showed a low MIB-1 labeling index (<3%). Adjacent colonic tissue showed areas of haemorrhage and focal denudation of overlying epithelium. There was no evidence of tumour in these areas. Mesenteric lymph nodes revealed reactive hyperplasia. Overall features were compatible with a well-differentiated liposarcoma involving the descending colon leading to a colo-colic intusussception.

Discussion

Liposarcomas are common soft tissue sarcomas which usually occur in the retroperitoneum and deep soft tissues of trunk and extremities. Although liposarcomas of the retroperitoneum can secondarily involve the gastrointestinal system, occurrence of primary liposarcomas in this region is uncommon and has been reported in the esophagus, stomach and small intestine. Primary colonic liposarcoma is extremely rare, and a search of literature revealed only six previous reports in the English literature [1–6]. The clinicopathologic features of these cases are summarised in Table 1.

Adult intussusception is a different entity from that found in children. An organic etiology is found in 90% of adult intussusceptions. Polypoid adenomas and lipomas are the most common benign leading points found in adult colo-colic intussusception, whilst colonic adenocarcinomas are most common malignant causes of adult intussusception.

In the few reported cases of primary colonic liposarcomas, the growth pattern was typically intraluminal as was seen in the present case. However, none of the cases reported thus far have presented as bowel obstruction. A palpable lump was seen in two patients, whilst one patient showed anaemia with significant weight loss. In the present case, also a palpable abdominal lump was noticed along with altered bowel habits and passage of blood and mucus in stool. In addition she was found to have a colo-colic intusussception due to the polypoidal intraluminal mass.

Radiological features of colonic liposarcomas are not well characterised owing to the small number of cases reported. Only one case where CT examination was performed showed a well-circumscribed homogenous mass showing low attenuation [5]. The preoperative evaluation of liposarcomas arising in colon is rendered even more difficult in cases where these lesions cause colonic intussusception as seen in the present case. Identification of the lead point of the obstruction and accurate characterisation of the lesion can pose a significant diagnostic challenge. In the present case, CECT of abdomen revealed left colo-colic intusussception with intramural mass lesion showing heterogenous contrast enhancement.

Table 1 Clinicopathologic features of reported cases of colonic liposarcoma

S no.	Age/sex	Clinical presentation	Location	Size (cm)	Histology	Follow-up	Ref.
1	62/F	Pain, palpable lump	Ileocaecal valve		Myxoid LPS	Died, 48 months	[1]
2	45/F	Diarrhoea, anemia, weight loss, abdominal discomfort	Caecum, Ascending colon	6X5X4	Pleomorphic LPS	NA	[2]
3	52/F	Pain, hematochezia	Descending colon	7.5×5.5×5	Well differentiated LPS	Alive, 24 months	[3]
4	46/M	Pain, palpable mass	Ascending colon	12×11×10	Myxoid LPS	Alive, 12 months	[4]
5	65/F	Pain, Obstruction	Caecum	5	Well differentiated LPS	Alive, 6 months	[5]
6	56/F	Hematochaezia, lower GI bleed	Hepatic flexure	3.5×3×2.8	Well Differentiated LPS	NA	[6]
7	66/F	Palpable lump, altered bowel habits, blood,mucus in stool	descending colon	4.5×3×2	Well Differentiated LPS	Alive, 10 months	Present case

Histologically, liposarcomas constitute a heterogenous group and according to the WHO classification are divided into five major histological subtypes: atypical lipomatous tumour (well-differentiated liposarcoma), myxoid liposarcoma, pleomorphic liposarcoma, dedifferentiated liposarcoma and mixed type liposarcoma. Among colonic liposarcomas, three cases of well-differentiated liposarcoma [3, 5, 6], two cases of myxoid liposarcoma [1, 4] and one case of pleomorphic liposarcoma [2] have been described in the literature. According to recent recommendations of the Working Group on Histological Classification of Soft Tissue and Bone Tumours, the term well-differentiated liposarcoma is reserved for deep-seated lesions arising in the retroperitoneum and mediastinum as compared to "atypical lipomatous tumour" for lesions with similar morphology occurring at surgically amenable locations [10]. Histologically, differentiation between lipoma and well-differentiated liposarcoma may be difficult and is based on the presence of atypia in the stroma and adipocytes. It is worth noting that identification of lipoblasts is not a prerequisite for the diagnosis of a well-differentiated liposarcoma. In fact, lipoblasts have been identified in several benign adipocytic tumours such as chondroid lipoma, lipoblastoma and pleomorphic lipoma [11]. In the present case also, no lipoblasts were identified, and the diagnosis was based on other histomorphological features.

Histological subtypes of liposarcomas have been shown to correlate with clinical behaviour. The well-differentiated subtype is considered to be a low-grade malignancy with a 5-year survival rate estimated between 75% and 100%, whilst the 5-year survival rates for pleomorphic and round cell liposarcoma are between 0% and 20% [10]. Although a long-term follow-up of patients with primary colonic liposarcoma is not available, most of the patients were symptom-free and clinically stable at the last follow-up [2–5]. There is only one report of a patient expiring 4 years after the initial diagnosis [1]. Our patient has been followed up for 6 months and is clinically stable with no evidence of recurrence or metastases at the last visit.

The protocol for treatment of liposarcoma of the colon has not been well established. However, most authors have recommended complete wide excision as the treatment of choice for such tumours [1, 2, 4].

Conclusion

In summary, the present case represents the seventh reported occurrence of primary colonic liposarcoma and the first presenting with a colo-colic intussusception. Although rare, primary colonic liposarcoma should be considered in the differential diagnosis of lesions in this area and particularly in cases which present with intussusception. A surgical exploration and histopathological examination to confirm the diagnosis and further management will thus continue be the gold standard in these cases.

References

1. Wood DL, Morgenstern L. Liposarcoma of ileocaecal valve: a case report. Mt Sinai J Med. 1989;56:62–4.
2. Parks RW, Mullan FJ, Kamel HM, Walsh MY, McKelvey ST. Liposarcoma of the colon. Ulster Med J. 1994;63:111–3.
3. Magro G, Gurrrera A, Di Cataldo A, Licata A, Vasquez. Well differentiated lipoma like liposarcoma of the caecum. Histopathology 2000;36(4):378–80. doi:10.1046/j.1365-2559.2000.0855d.x.
4. Chen KT. Liposarcoma of the colon: a case report. Int J Surg Pathol. 2004;12:281–5. doi:10.1177/106689690401200312.
5. Gutsu E, Ghidirim G, Gagauz I, Mishin I, Iakovleva I. Liposarcoma of the colon: a case report and review of the literature. J Gastrointest Surg. 2006;10:652–56. doi:10.1016/j.gassur.2005.09.014.
6. Shahidzadeh R, Ponce CR, Lee JR, Chamberlain SM. Liposarcoma in a colonic polyp: case report and review of the literature. Dig Dis Sci. 2007;52(12):3377–80. doi:10.1007/s10620-007-9806-4.
7. Takeuchi K, Tsuzuki Y, Ando T, Sekihara M, Hara T, Kori T, et al. The diagnosis and treatment of adult intussusception. J Clin Gastroenterol. 2003;36:18–21. doi:10.1097/00004836-200301000-00007.
8. Goh BK, Quah HM, Chow PK, Tan KY, Tay KH, Eu KW, et al. Predictive features of malignancy in adults with intussusception. World J Surg. 2006;30:1300–4. doi:10.1007/s00268-005-0491-1.
9. Kim T, Murakami T, Oi H, Tsuda K, Matsushita M, Tomoda K, Fukuda H, Nakamura H. CT and MR imaging of abdominal liposarcoma. Am J Roentgenol 1996;166:829–33.
10. Christopher DM, Unni KK, Mertens F. Liposarcoma. WHO classification of tumors. Pathology and genetics: tumors of soft tissue and bone. Lyon, France: IARC; 2002. p. 35–46.
11. Meis JM, Enzinger FM. Chondroid lipoma; a unique tumour simulating liposarcoma and myxoid chondrosarcoma. Am J Surg Pathol. 1993;17:1103–12. doi:10.1097/00000478-199311000-00003.