

# Fistulation as a complication of intra-abdominal soft-tissue sarcomas; a case series

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## ABSTRACT

Soft-tissue sarcomas are rare, accounting for only one percent of all cancers. They can occur in retroperitoneal and intraperitoneal sites, including gastrointestinal stromal tumours (GIST), and have the potential to cause complications secondary to interaction with other abdominal viscera. Fistulation, or an abnormal communication between two epithelium-lined surfaces that do not usually connect, is a rare example of such a complication. We present a series of cases of fistulation due to the presence of an intra-abdominal soft-tissue sarcoma and contrast three different approaches to management. We discuss the radiological features and other modalities of imaging which may be useful in diagnosing this rare complication.

## CASE SERIES

### CASE SERIES

#### *Case One*

A 73-year-old female presented with a three day history of vomiting, abdominal distension and constipation. She had a history of a well-differentiated retroperitoneal liposarcoma that was resected along with the right kidney four years previously. Four months prior to this presentation, a follow-up staging computed tomography (CT) had demonstrated recurrent disease with an 18 x 21cm retroperitoneal mass which was deemed inoperable and she was offered palliative chemotherapy [figure 1]. Following the first cycle of chemotherapy, the patient was admitted with vomiting and found to have bowel obstruction secondary to extrinsic compression of the duodenum by the tumour. She was managed with endoscopic insertion of a duodenal stent and the symptoms resolved.

On this presentation, the symptoms were similar suggesting recurrent bowel obstruction along with fever and signs of sepsis. Physical examination revealed a large right-sided abdominal mass that was tender to palpation and

surprisingly, tympanic to percussion. CT of the abdomen with intravenous contrast demonstrated a heterogeneous retroperitoneal mass composed of soft tissue and fat as seen previously, but with new multiloculated gas within the anterior component suggesting a communication with the bowel lumen. Furthermore, there was radiographic suggestion of duodenal stent erosion at its proximal margin with a duodenal fistula accounting for the new appearances. The first and second parts of the duodenum appeared effaced due to mass effect, accounting for the patient's symptoms of outflow obstruction [figure 2].

The patient was made nil by mouth, a nasogastric tube was inserted for drainage, and she was treated with intravenous antibiotics to cover for intra-abdominal sepsis. She underwent oesophagogastroduodenoscopy (OGD) where a covered metal duodenal stent was inserted within the lumen of the previous stent. This effectively covered the site of the duodenal fistula tract and relieved the duodenal compression. Soon after endoscopic intervention the patient improved with complete resolution of symptoms of bowel obstruction. She was able to tolerate increasing amounts of oral diet and was eventually

discharged. Unfortunately due to poor performance status, she was not offered any further chemotherapy and the patient passed away four months after this presentation.

### Case Two

A 37-year-old female with metastatic synovial sarcoma presented with a one week history of vomiting with haematemesis, anorexia and lethargy. She had recently completed a course of palliative chemotherapy for disease progression in the liver, bone and a left-sided retroperitoneal mass. On examination, the patient was clinically anaemic with a mildly distended and tender abdomen and a palpable left-sided abdominal mass. Upper gastrointestinal haemorrhage was suspected and the patient proceeded to have an OGD. This showed an infiltrating mass in the second part of the duodenum with evidence of bleeding. An urgent contrast CT of the abdomen confirmed further disease progression with the retroperitoneal mass now invading into the second part of the duodenum. There were also new appearances of multiple locules of air within compatible with fistulation. There was noted to be marked gastric and bowel dilatation to the level of the second part of the duodenum where the mass appeared to fistulate with bowel [figure 3].

The patient was initially managed with blood transfusion, intravenous fluid resuscitation, decompression of the stomach with nasogastric tube insertion and intravenous antibiotics. Endoscopic management of the obstruction and fistulation was considered, however it was felt that the extent of obstruction in this case was not amenable to stenting. A surgical review was sought and the patient proceeded to have palliative surgery with laparotomy and formation of a gastro-jejunostomy to bypass the obstruction. Post-operatively the patient improved steadily and was able to resume oral intake. She was discharged from hospital after a three-week admission and is now undergoing second line palliative chemotherapy.

### Case Three

A 71-year-old male with a gastro-intestinal stromal tumour (GIST) of the stomach and oesophagus presented with ongoing abdominal pain, distension and a chronic haemoserous discharge from sinus on the anterior abdominal wall. A staging CT with contrast showed a partly cystic, partly solid exophytic mass arising from the greater curve of the stomach and communicating with its lumen, as well as a fistulous connection between the skin surface and the upper gastrointestinal tract [figure 4]. This fistula tract was best visualized on curved planar reformatted axial CT images and measured to be 8cm in length [figure 4b].

The patient failed to respond to conventional first and second line therapy with receptor tyrosine kinase inhibitors, Imatinib and Sunitinib. A surgical resection was attempted but abandoned due to the large size of the tumour and intraoperative haemodynamic instability. The patient deteriorated over a period of months, with worsening abdominal pain, regular passage of melaena necessitating hospital admission for large volume blood transfusion, and worsening fistulous discharge from the anterior abdominal wall significantly impacting on quality of life. The patient's complicated case was discussed at a regional sarcoma multi-

disciplinary meeting and a recommendation made for palliative radiotherapy. This was administered as a single 10Gy fraction to the mass.

On review eight weeks later, the patient had had a remarkable response with complete cessation of symptoms of melaena and stable haemoglobin within normal range. Furthermore there was complete resolution of discharge from fistula site. A repeat staging CT confirmed regression of the fistula tract, along with a decrease in the size of the mass associated with the stomach [figure 5]. At the time of writing, nearly 10 months after delivery of radiotherapy, the patient continues to be well on regular follow-up.

## DISCUSSION

Soft-tissue sarcomas are rare and account for around only one percent of all cancers in adults [1]. Retroperitoneal and intraperitoneal sites, including gastrointestinal stromal tumours (GIST), make up twenty percent of soft-tissue sarcomas [2]. These usually present with pain or a gradually enlarging mass, but can be complicated by local pressure effects such as bowel obstruction or hydronephrosis. Fistulation as a complication of intra-abdominal soft-tissue sarcoma is rare. Only one other case report has been found, where a recurrent retroperitoneal liposarcoma fistulated with the descending colon [3]. Fistulas in general occur either as a complication of abdominal surgery, or spontaneously as a result of intra-abdominal inflammation or infection. In the context of malignancy they are more commonly reported in adenocarcinoma of the colon resulting in gastrocolic fistulas [4,5,6].

In this case series we have demonstrated that fistulation can occur in a range of soft-tissue sarcoma histological subtypes, and was a complication of progressive or recurrent disease in all three cases. Aetiology is unknown, but may be related to the large disease bulk that is often seen. Clinical presentation is variable depending on the site of fistulation; involvement of the gastrointestinal tract leads to symptoms such as vomiting, which have a broad differential diagnosis. Imaging is therefore an important part of the diagnostic work-up.

### Imaging

Due to its speed, ease of access and reproducibility, CT scanning is the 'workhorse' of oncology imaging. Radiological features of fistulation depend on the intra-abdominal organ with which a tumour is communicating and should always be compared with clinical findings. Although CT has very good spatial resolution, inherent technical limitations can fail to resolve small focal ingrowth with a solid organ of similar soft tissue density. Similarly, the actual fistulous tract through the soft tissue wall of a hollow viscus can appear covert at imaging. It is often the sequelae of communication that allows for a radiological diagnosis of fistulation.

Exophytic growth into a hollow viscus (for example the duodenum) allows air to move into the communicating tumour and coalesce between fascial planes. The disparity in density

between soft tissue and air produces characteristic CT appearances as demonstrated in Figure 2. In this context, CT imaging is the modality of choice, as other modalities including Ultrasound (US) and Magnetic Resonance Imaging (MRI) would be unable to accurately resolve locules of gas. Although the clinical picture along with CT findings confirms the diagnosis, possible differentials include infection with gas-forming organisms, perforation of abdominal viscera or post-surgical or post-intervention appearances.

MRI can play an important role in determining fistulation between a tumour and soft tissue organ of similar density, for example within the female pelvis. T2 weighted imaging would have an anatomical advantage over CT when identifying a fistulous tract. Its superior contrast scale when resolving soft tissue can allow for highly accurate characterisation to aid the clinical team in management of such patients. Ultrasound offers limited use in the diagnosis of fistulation and often only provides anatomical information when the fistula is superficial or communicating with the skin surface. In conjunction with another imaging modality, it may offer some use in the follow-up of a fistula or in its management. Similarly, the use of fluoroscopy can be very helpful when trying to characterise the extent of a fistulous tract.

#### Management

Being a rarely reported phenomenon, there are no guidelines or literature to recommend the best way of managing fistulation in soft-tissue sarcomas. Management of intra-abdominal fistulas in general is a clinical problem most commonly encountered in the context of inflammatory bowel disease. It is widely accepted that the mainstay of management there is surgical, with resection of the fistula tract along with the associated segment of bowel [7]. Other important considerations include fluid or blood resuscitation, considering parenteral nutrition and avoidance or treatment of infection. These conservative measures are also relevant in the management of fistulation in soft-tissue sarcomas, and were key in the management of the cases in this series.

Unfortunately, surgical resection of the fistula tract and associated viscera is rarely possible in the context of soft-tissue sarcomas. The intra-abdominal disease bulk can often be large, and patients can have high peri-operative risk due to comorbidities and the physiological burden of cancer. In the second case, the patient was a 37-year-old female who was otherwise fit, making her a good candidate for successful bypass surgery. We have, however, demonstrated other non-surgical approaches with endoscopic stenting and radiotherapy, which also resulted in a positive outcome.

Endoscopic stenting with covered stents is routinely used for tracheo-oesophageal fistulas, which are often secondary to malignancy [8]. They are also recommended in management of obstructing colonic malignancies, with covered stents being particularly useful when there is an associated enteroenteric fistula [9]. Radiotherapy is not routinely used to treat enteric fistulation and indeed nor is it routine in the management of GIST. In the third case, radiotherapy was offered as neither surgery nor standard targeted therapy was successful. It

resulted in control of tumour progression and also of associated fistulation and haemorrhage.

#### Conclusion

In a large tertiary referral oncology unit with a dedicated sarcoma service, fistulation in soft-tissue sarcoma may be a significant complication that needs to be considered. We have presented three cases with different clinical features on presentation. We have highlighted the radiological characteristics of fistulation and other imaging modalities which may be diagnostically helpful, and contrasted three different approaches to management. In each case, decisions regarding management were made in a multidisciplinary setting on an individual basis, ensuring the best outcome for the patient.

#### TEACHING POINT

Fistulation as a rare complication of soft-tissue sarcoma is best diagnosed on CT and key features suggesting fistulation include the appearance of air within a tumour and identification of the fistula tract itself. Management strategies can be varied, including surgical, endoscopic and radiotherapy, and is best undertaken in a multidisciplinary setting at a dedicated sarcoma oncology unit.

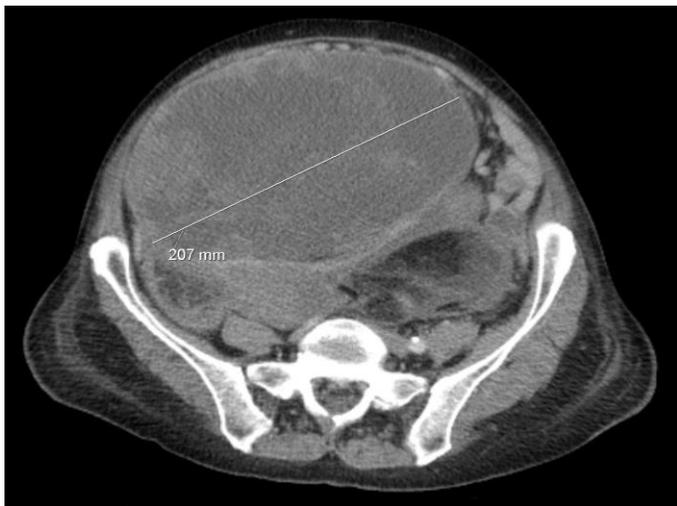
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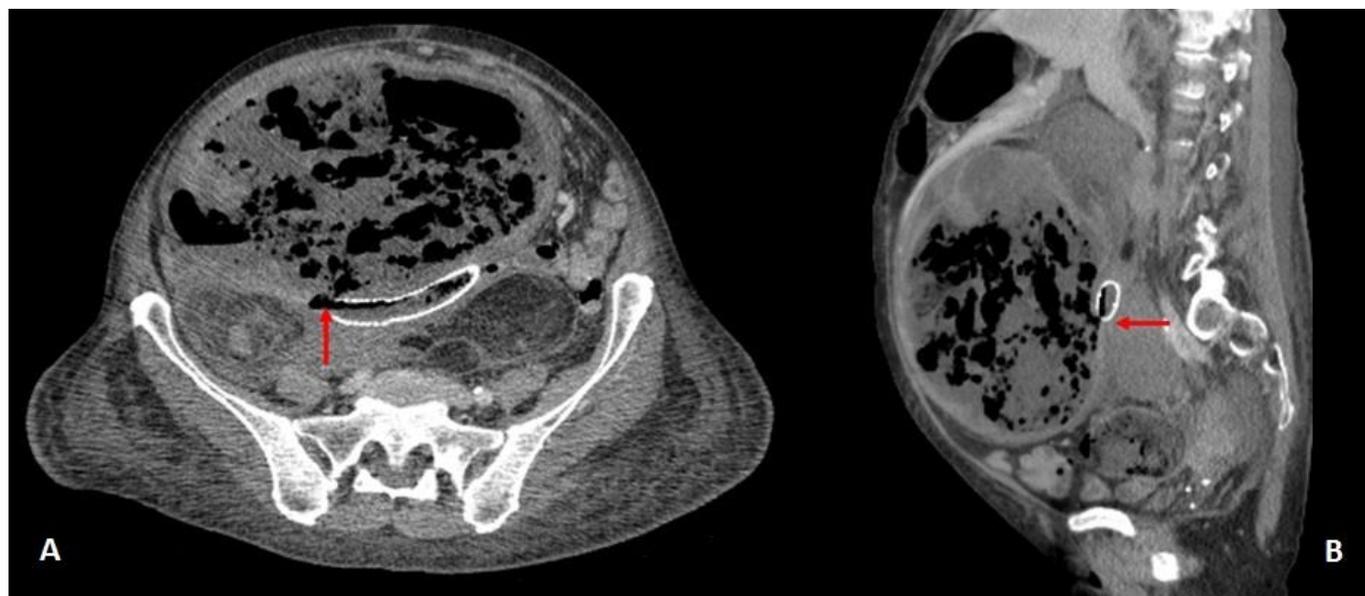
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FIGURES



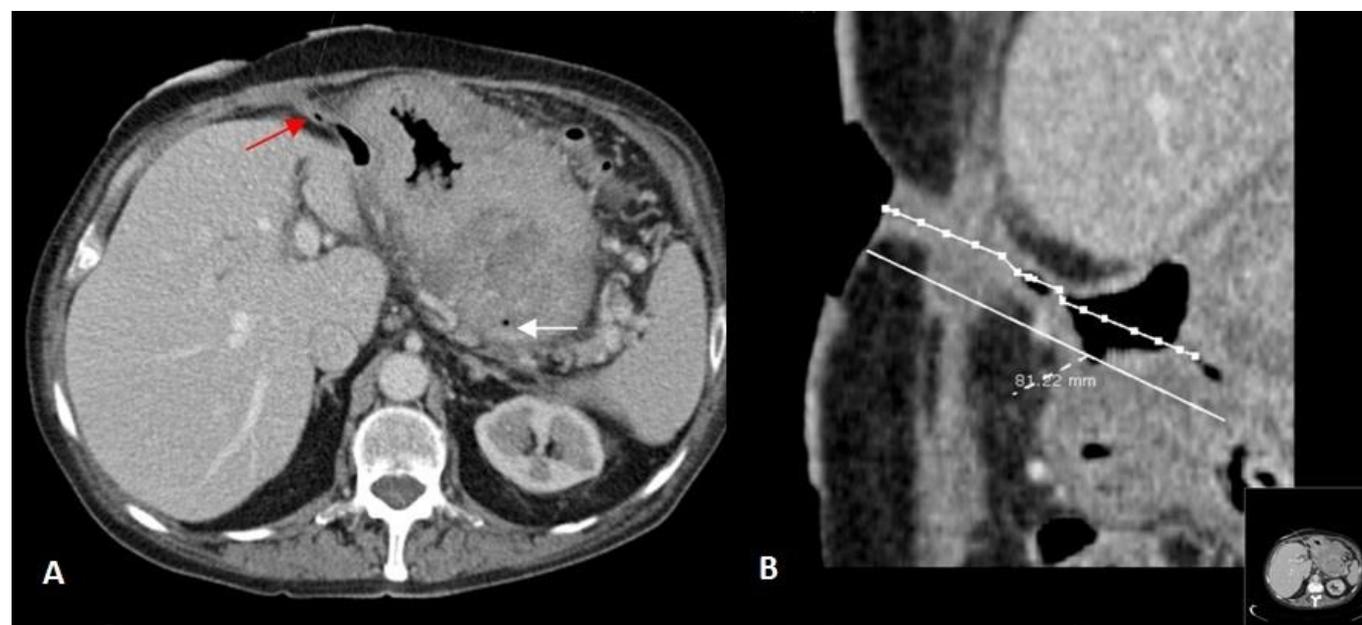
**Figure 1 (left):** 73-year-old female with recurrent retroperitoneal liposarcoma. Axial staging CT image obtained in portal venous phase (GE Lightspeed Pro 32, 1.25mm slice thickness, 120kVp, 200mAs) with intravenous contrast (100ml Omnipaque 300) demonstrates a large heterogeneous mass composed of soft tissue and fat.



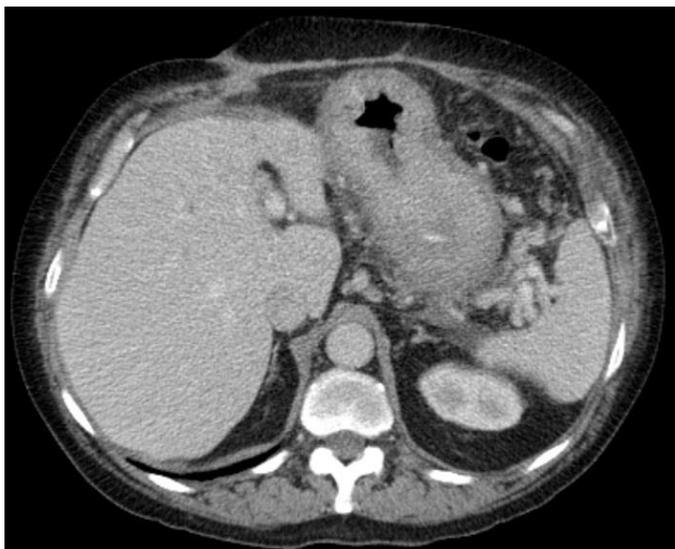
**Figure 2:** 73-year-old female with recurrent retroperitoneal liposarcoma presenting with vomiting following a recent episode of bowel obstruction due to tumour compression of the duodenum which was treated with endoscopic stenting. Axial (A) and sagittal (B) CT images in the portal venous phase (GE Lightspeed Pro 32, 1.25mm slice thickness, 120kVp, 200mAs) with intravenous contrast (100ml Omnipaque 300) demonstrate multiloculated gas within the anterior component of the mass. The previous duodenal stent is visualised with an associated duodenal fistula (arrows).



**Figure 3:** 37-year-old female with metastatic synovial sarcoma presenting with vomiting and anorexia. Coronal CT of the chest, abdomen and pelvis (a) obtained in portal venous phase (GE Lightspeed Pro 32, 1.25mm slice thickness, 120kVp, 200mAs) with intravenous contrast (100ml Omnipaque 300) demonstrates a retroperitoneal mass invading into and fistulating with the second part of the duodenum (white arrow), containing locules of air (red arrow). Axial CT (b) further demonstrating a retroperitoneal mass with multiple locules of air (black arrow).



**Figure 4:** 71 year-old-man with gastrointestinal stromal tumour (GIST) of the oesophagus and stomach presenting with abdominal pain and distension, and a haemoserous discharging sinus on the anterior abdominal wall. Axial CT (A) with contrast (Ultravist 300) in the portal venous phase (Siemens sensations 63, 1.5mm slice thickness, 120mVp, 200mAs) demonstrates an exophytic mass arising from the greater curve of the stomach and containing locules of air (white arrow), suggesting a communication with the lumen of the stomach. There is also a fistulous connection between the skin, the tumour and the upper gastrointestinal tract (red arrow). This is better seen on curved planar reformatted axial CT images (B) which shows an 8cm fistula tract.



**Figure 5:** 71 year-old-man with gastrointestinal stromal tumour (GIST) of the oesophagus and stomach complicated by a fistula between the skin surface and upper gastrointestinal tract. Post-radiotherapy axial CT with contrast (Ultravist 300) in the portal venous phase (Siemens sensations 63, 1.5mm slice thickness, 120mVp, 200mAs) demonstrates resolution of the fistula tract and decrease in the size of the soft-tissue mass associated with the stomach. Absence of intra-lesional free gas also suggests resolution of the fistula.

<b>Etiology</b>	Slow-growing, large intra-abdominal disease bulk with interaction and invasion into surrounding viscera
<b>Incidence</b>	Estimated at <1% of soft-tissue sarcoma
<b>Gender ratio</b>	Uterine leiomyosarcoma a major subtype of soft-tissue sarcoma, therefore F > M
<b>Age predilection</b>	Fistulation = a complication of advanced or progressive disease, therefore may be seen more in older adults
<b>Risk factors</b>	Large intra-abdominal disease bulk, progressive disease, treatment resistance
<b>Treatment</b>	Surgical resection; bypass surgery; endoscopic stenting; radiotherapy; chemotherapy/targeted therapy for underlying disease
<b>Prognosis</b>	Poor as tends to be associated with advanced or progressive disease
<b>Findings on imaging</b>	CT – air within the tumour, identification of a fistula tract, associated signs of bowel obstruction

**Table 1:** Summary table of fistulation in soft-tissue sarcoma

Differential	CT	Clinical features
<b>Fistula</b>	Air within solid mass; communication/close interaction with bowel; visible fistula tract	Depends on site of fistulation – signs of obstruction, intra-abdominal sepsis, visible discharge
<b>Infection with gas-forming organism</b>	Air within a solid mass/organ; associated collections and fat-stranding	Clinically septic, positive blood cultures (e.g. Clostridium perfringens)
<b>Perforation</b>	Free air outside of bowel/abdominal mass	Peritonism, vomiting, bowel obstruction/ileus
<b>Post-surgical</b>	Intra-peritoneal free air	Positive history
<b>Post-intervention (e.g. biopsy, drainage)</b>	Intra-peritoneal free air, or air within mass	Positive history

**Table 2:** Differential diagnosis table of fistulation in soft-tissue sarcoma

**ABBREVIATIONS**

CT = computed tomography  
 GIST = gastrointestinal stromal tumour  
 MRI = magnetic resonance imaging  
 OGD = oesophagogastroduodenoscopy  
 US = ultrasound

**KEYWORDS**

Soft-tissue sarcoma; Fistula; CT

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