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Case report

Mesenteric cyst with a twist

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Abstract

With an incidence rate of 0.1-0.3% gastrointestinal stromal tumors (GIST) are rare to be found in extraintestinal locations known as extraintestinal GIST (EGIST). Mesenchymal in origin, they arise from interstitial cells of Cajal, a part of gastrointestinal autonomic nervous system (ANS). It is more common in males than females. GIST is mainly seen in stomach followed by small bowel, colon, rectum and esophagus. Incidence of EGIST is less than 1%. Patients with neurofibromatosis-1 has increased incidence of GIST. It can also present as Carney-Stratakis syndrome. GIST less than 2 cm is usually asymptomatic while GIST more than 2 cm presents with unexplained growth of abdomen, nausea, anemia, difficulty in swallowing, loss of appetite and weight loss. Herein we present a case of 31-yr old female, who presented with complaints of intermittent abdominal pain and was evaluated for infertility. Abdominal ultrasonography (USG) revealed cystic mass of mesentery. Computed tomography (CT) scan abdomen confirmed a cystic lesion with central necrosis arising from jejunal loops. Surgical excision of mesenteric cyst was performed and histopathological diagnosis of gastrointestinal tumor was given. Thus, high index of suspicion is required on part of pathologist to expect an EGIST mimicking as a mesenteric cyst.

Key words: Extra gastrointestinal stromal tumor, mesenteric cyst, surgical resection

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Gastrointestinal stromal tumors are mesenchymal tumors of gastrointestinal tract which arises from interstitial cells of Cajal within myenteric plexus of the muscularis propria[1].The term GIST was first used by Mazur and Clark in year 1983[2]. GIST overexpresses tyrosine kinase receptors mainly due to mutation in CD117 (C-kit) or platelet derived growth factor receptor alpha (PDGFRA)[1]. Incidence of gastrointestinal tumor is 60% in stomach, 20-30% in small bowel,

5% in colon and rectum, 5% in esophagus and less than 1% in other sites like mesentery and omentum[1,3]. Extraintestinal GIST may be metastasis from GIST or a subtype of GIST. Patient is asymptomatic initially and later presents with complaints of abdominal pain, bleeding due to hyper vascular nature of GIST, abdominal mass. Endophytic growth in muscularis propria of intestine presents as intestinal obstruction or bleeding whereas exophytic growth presents as a mass lesion or intestinal perforation[3]. GIST is more common in age of 50 years or above and there is no gender predilection[1]. When complete surgical resection is done there is better chance of survival[1].Imatinib which is tyrosine kinase receptor inhibitor has a major response postoperatively as GIST lack response to radiotherapy and chemotherapy[4].

Case

This case presented in Gynecology department of Princess Esra Hospital was a 31-year-old female who had complaints of intermittent abdominal pain and was evaluated for infertility.

On investigations, Complete Blood Picture (CBP) revealed neutrophilic leucocytosis. Ultrasonographic examination revealed thickened mass with unilocular cystic cavity. Contrast enhanced computed tomography (CECT) revealed well-circumscribed areas in abdominal cavity showing homogenous peripheral enhancement receiving blood supply from superior mesenteric artery and central non-enhancing hypodense fluid in close proximation to ileal loops showing claw sign positive hinting of GIST with central necrosis (Fig 1).

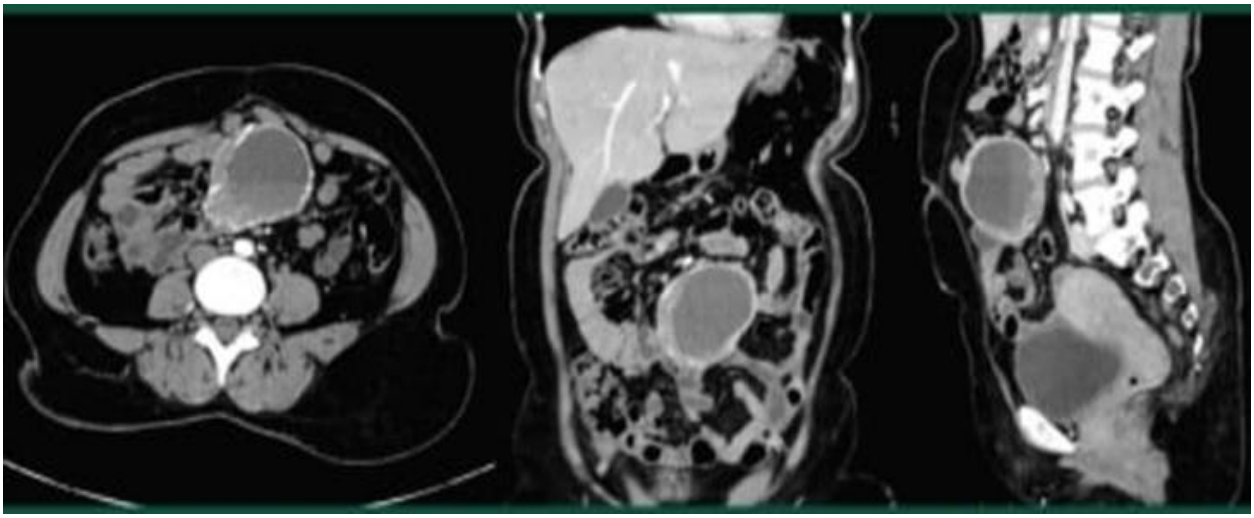

Fig 1. CECT image showing well circumscribed mass, with claw sign

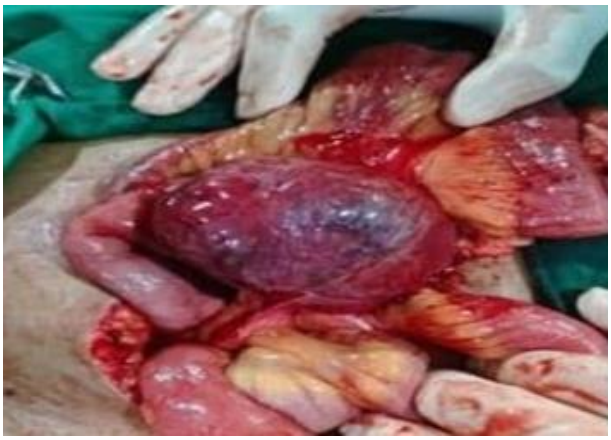

Fig 2. Intra-operatively noted mesenteric cyst

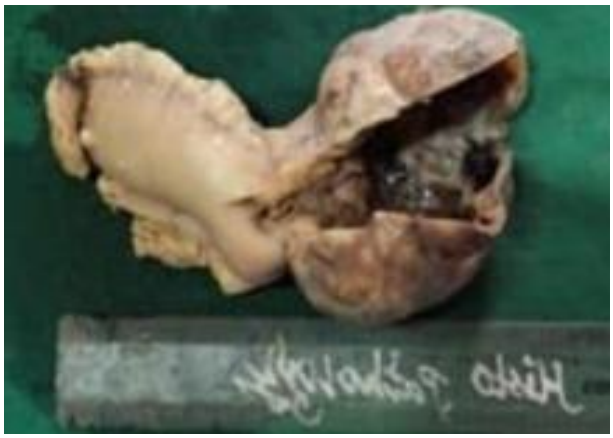

Fig 3. Excised abdominal mass

The patient was operated. Intraoperatively mesenteric cyst was noted measuring 7x7cm, 10cm distal to duodeno-ileal junction arising from jejunal loops (Fig 2). The resected tissue was received in formal saline. Gross examination showed cystic mass.

External surface showed glistening, smooth surface. Cut-section ascertained unilocular, greyish white solid mass measuring 5x3 cm. It was found attached to small intestine (jejunum) measuring 7 cm (Fig 3).

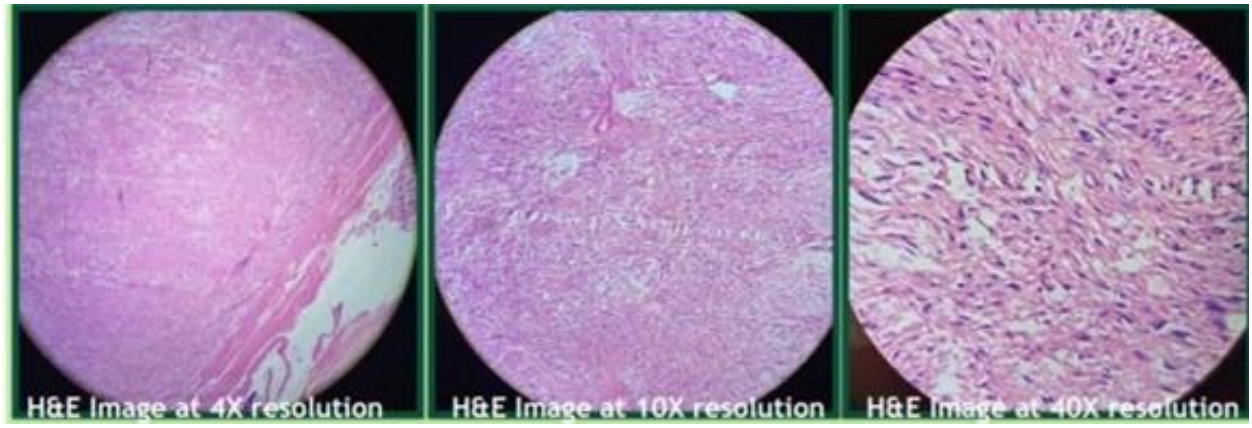

Fig 4. Histopathological slides of GIST

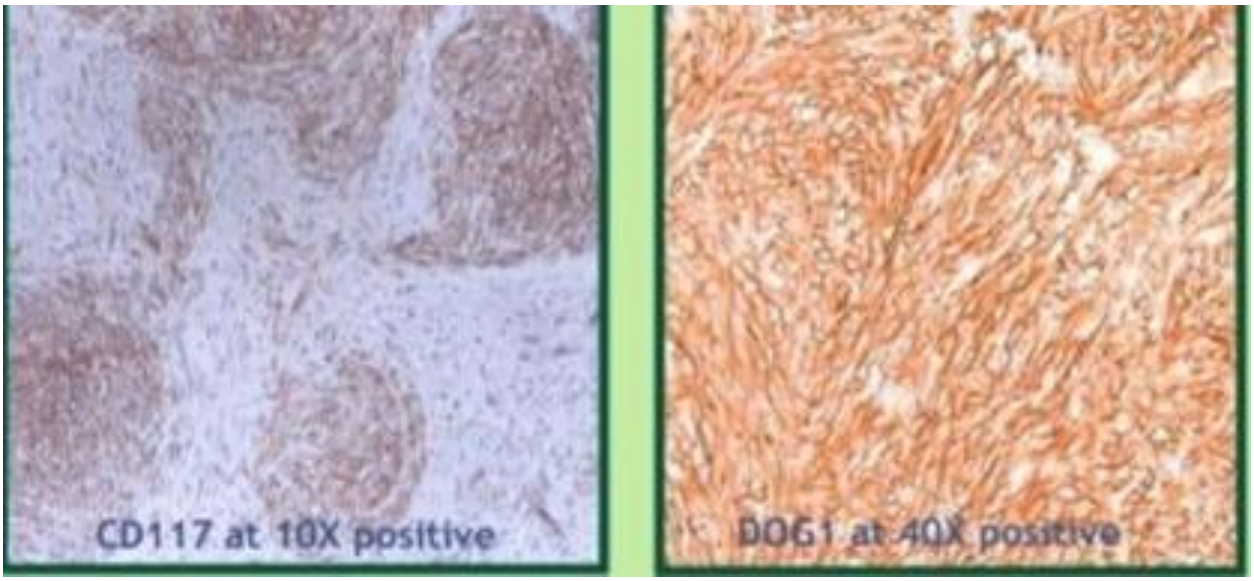

Fig 5. IHC positive

On microscopy there was no lining epithelium in the cystic mass and stroma mainly consists of spindle shaped cells constituting about 70%, epitheloid cells constituting 20% and mixed type constituting about 10%, nucleus being pleomorphic with eosinophilic cytoplasm. Scanty mitosis identified. No evidence necrosis.

Immunohistochemistry (IHC) examination was positive for CD117 and discovered on GIST 1 (DOG-1). Based on the results the diagnosis of EGIST was made.

Discussion

GIST is common among 3rd to 4th decades of life from above information which is also seen in our case. From above information GIST is common in males unlike in our case who is female. The main presenting symptom is abdominal pain also seen in our case but can also present with abdominal distension, vomiting and also palpable mass. GIST is a rare entity which was previously not differentiated from other non-epithelial neoplasm[3].The term GIST was first used by Mazur and Clark in 1983[1,2]. GIST are defined as spindle, epitheloid and occasionally pleomorphic mesenchymal tumor of gastrointestinal tract expressing kit protein[3]. GIST is rare entity comprising of 0.1 – 0.3 % of gastric neoplasm[3]. EGIST is a subset of GIST occurring outside gastrointestinal tract. Histopathologically and immunohistochemically EGIST resemble GIST[3]. Due to mutation in ckit and PDGFRA there is activation of tyrosine kinase receptors leading to uncontrolled cell proliferation and survival[1]. GISTs show broad spectrum of differentiation ranging from myoid, neural, dual differentiation to undifferentiated tumor[3]. Presentation of GIST varies depending on the size of tumor and its anatomical location, complications[3]. There is increased incidence of GIST in patients with neurofibromatosis type-1. Small GIST which are often asymptomatic are usually seen as incidental finding radiologically or endoscopically.

Larger GIST presents with non-specific symptoms. In our case the complaint of patient was abdominal pain presented as mesenteric mass arising from small intestine then by microscopy confirmed it as EGIST. Predominant cell type on microscopy is spindle shaped cells[3]. In cases of abdominal pain ultrasonography can be done as it provides initial formation but is limited to its specificity revealed cystic unilocular mass[1].CECT provides a detailed and specific information showing peripheral enhancement with non-enhancing hypodense fluid showing claw sign positive. This sign is shown when tumor extends into surrounding normal tissue creating claw like appearance on imaging. Final diagnosis of GIST can only be confirmed by CD117 and DOG-1[3]. In our case IHC revealed that patient has chances of malignant potential and chances of recurrences. Prognosis of GIST mainly depends on tumor size, anatomical location, mitotic rate[3].

Primary treatment for GIST is complete surgical excision of tumor[3,4]. Recurrence and metastasis can be prevented by complete resection[1]. In our case surgical excision followed by resection and anastomosis of jejunal loops was done. Tumors measuring more than 5 cm and with high mitotic rate have a high chance of recurrence and metastasis. For patients at a higher risk imatinib therapy is recommended which is a tyrosine kinase receptor inhibitor thereby inhibiting cell proliferation and survival[4].

Conclusion

As mesenteric GIST is a rare entity it can be clinically mistaken for mesenteric cyst. Emphasizing the role of radiological examination and pathological investigation to arrive at a final diagnosis. Despite rareness of EGIST it should be considered in differential diagnosis of abdominal pain for treatment in appropriate time and for decreasing chances of recurrences. Also underscores the importance of immunohistochemistry in confirmation of final diagnosis and assessing malignant potential of tumor. GIST is common in age of 50 years or above with no gender predeliction. Thus, GIST should be considered in differential diagnosis of abdominal mass in younger females.

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Conflict of interest: None

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