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CASE REPORT

Meckel's Diverticulum (Gastrointestinal Stromal Tumors)-Two Rare Case Reports

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ABSTRACT

Introduction: Meckel's Diverticulum is the most common congenital anomaly of the gastrointestinal tract (incidence: 0.6%-4%). Gastro Intestinal Stromal Tumors originate from multipotential stromal cells of Cajal (pacemaker cell of gastrointestinal tract). The GISTs of Meckel's diverticulum are extremely rare,especially if you consider that only 2% of the population has this kind of diverticula confirmed by autopsy studies, with a percentage that varies between 0.4 and 4.5%.

Case Reports: We report two cases, a 53 year old post menopausal lady and a 45 year old peri menopausal lady, both clinico radiologically diagnosed as ovarian masses, found to have a tumor arising from Meckel's Diverticulum intraoperatively, that were histopathologically proven to be Gastro Intestinal Stromal Tumors. A thorough literature review was done and all the previously quoted case reports are listed. Our literature search revealed a total of 42 case reports of GIST of Meckel's Diverticulum including ours. Perforation is the most common presentation of a Meckel's GIST (13 case reports).Meckel's GIST presenting as a pelvic mass mimicking an ovarian pathology has been reported in the literature (6 case reports).

Conclusion: Meckel's diverticular tumours, though rare, may mimic other pathologies of the abdomen, therefore should be taken into consideration during the differential diagnosis of abdominal and pelvic tumours, especially in cases where imaging modalities doesn't pinpoint adefinitive diagnosis.

INTRODUCTION

Gastrointestinal stromal tumors (GISTs) are commonest mesenchymal tumors of the gastrointestinal tract, accounting for 0.1%-1% of gastrointestinal malignancies, having distinct biological properties and characterized by CD117 expression on IHC with stomach being the commonest anatomical location.[1,2] Meckel's Diverticulum (MD), a congenital abnormality of the gastrointestinal tract occasionally can harbour GIST. The reported incidence of tumors in Meckel's diverticulum is 0.5 to 3.2%.[3] To our knowledge, 40 cases have been previously reported. The current case series is unique in the way they presented and lead to significant clinical diagnostic challenge.

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CASE REPORT 1

A 53 year old postmenopausal lady presented to the gynecology department with vague lower abdominal pain since 3 months and no other complaints. Per abdominal examination revealed no abnormality, per vaginal examination revealed a hard mass palpable in the pouch of Douglas, not moving with the uterus. Rest of physical examination was unremarkable. Ultra sonogram of abdomen and pelvis showed a heterogenous mass of size 8x6cm in the pelvis without a clear demarcation between the right ovary and the tumor. She was referred to us for further management. Contrast Enhanced Computed Tomography (CECT) abdomen and pelvis revealed a lobulated mass lesion in the pelvis posterior to the uterus and the right ovary could not be made out separately from the tumour with a probable diagnosis of right ovarian mass (Figure 1a). Ovarian tumor markers and Serum CEA were within normal limits.

Primary cytoreduction was planned and lower midline laparotomy was performed. On laparotomy, a tumor

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of size 8x6 cm is found in the pelvis posterior to the uterus with its origin from a short diverticulum of the ileum at about 50 cm from the ileocaecal junction (Figure 1b). Hence it was diagnosed as a tumor arising from the Meckel's Diverticulum. The right ovary is normal but is densely adherent to the tumor and hence right oophorectomy was done. Rest of abdomen was normal. Tumour was excised with 2 cm of ileum on either side. End to end anastomosis was done in single layer. Post operative period was uneventful and the patient was discharged on 5th post operative day.

The Histopathology of the tumor was reported as Gastrointestinal Stromal Tumor arising from the Meckel's Diverticulum. The tumour showed spindle cells in fascicles (Figure 1c). All margins were negative. It was a low grade tumor with mitotic rate <5/50 HPF. Immuno-hisochemistry showed positive reaction for CD117 (C-Kit) and DOG1 (Figure 1d-e). But desmin, actin, S100, and CD 34 were negative. The final stage of the tumor is T3 N0 M0 G1- stage IB. Patient was planned for adjuvant Imatinib therapy.

CASE REPORT 2

A 45 year old perimenopausal lady presented to the gynaecology department with irregular menstrual cycles and vague lower abdominal pain occasionally since 6 months and no other complaints. Per abdominal, per vaginal and per rectal examination revealed no abnormality and a routine Ultra sonogram abdomen and pelvis revealed a complex mass in the pelvis about 4x3 cm not separately made out from the right ovary and hence was referred to us for further management.

CECT abdomen and pelvis revealed a hypoechoeic solid highly vascular focal lesion measuring 4.2 x3cm in the right adnexa related anteriorly to the right iliac vessels with the right ovary not seen separately from the tumor. Ovarian tumor markers and serum CEA are within normal limits.

Exploratory laparotomy was done and the tumor was found to be arising from the distal ileum and is adherent to the right ovary (Figure 2). Tumour was excised with 2 cm of ileum on either side. End to end anastomosis was done in single layer. Post operative period was uneventful and the patient was discharged on 4th post operative day.

The histopathology report suggested as Gastro Intestinal Stromal Tumor arising from the Meckel's Diverticulum. It was a low grade tumor with mitotic rate <5/50 HPF. Immuno-hisochemistry showed positive reaction for CD117 (C-Kit) and DOG1. The final stage of the tumor was T2 N0 M0 G1-stage IA. She was sent for medical oncologist consultation.

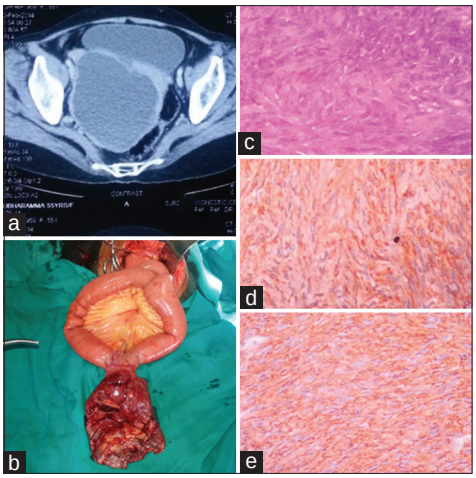

Figure 1: (a) Contrast enhanced CT Pelvis images showing lobulated mass lesion in the pelvis posterior to the uterus and the right ovary could not be made out separately from the tumour. (b) Intraoperative image showing a huge mass arising from the anti-mesentric border of the terminal ileum. (c) 40X, H&E staining, showing spindle cells in fascicles. (d) IHC- CD117 (C-Kit), Diffusely positive, 40X. (e) IHC- DOG1 Diffusely positive, 40X

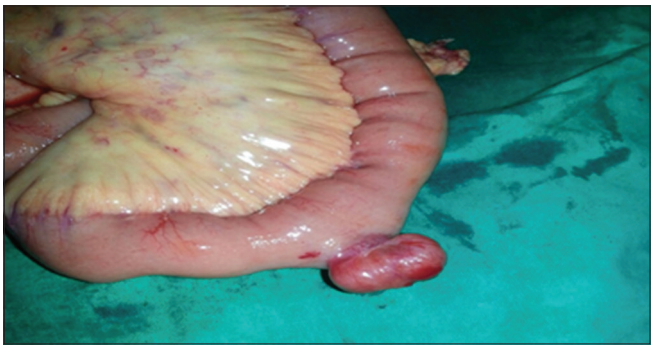

Figure 2: Intra operative image showing a tumor arising from the anti mesentric border of the terminal ileum

DISCUSSION

Meckel's Diverticulum is the most common congenital anomaly of the gastrointestinal tract (incidence: 0.6%-4%).[4,5] It is clinically insignificant unless complications occur. Only 4%-16% of Meckel's diverticula produce symptoms mainly caused by intussusception and ulceration. A neoplasm is a rare complication observed in only 0.5%-3.2% of Meckel's diverticula. Carcinoid tumors are the most common (33%-44%) primary diverticular malignancies, followed by leiomyosarcoma (18%-25%) and adenocarcinoma (12%-16%).[6,7]

Simple transverse resection is not recommended for the short Meckel's diverticulum. A height to diameter ratio (HDR) of 2.0 is recommended as the cut-off when deciding on the most appropriate operation. The external appearance of the Meckel's diverticulum does not predict the presence of heterotopic gastric mucosa and is therefore an unreliable indicator to aid resection decisions when presented with an incidental Meckel's diverticulum.[8]

GIST is a cancer of the gastrointestinal tract that originates from multipotential stromal cells of Cajal (pacemaker cell of gastrointestinal tract) that can develop and differentiate into smooth muscle cells or neurons that control gastrointestinal motility.[9,10] These tumours represent a percentage of 1% of all the gastrointestinal system cancers, with localization in the stomach (70%) and the small intestine (25%), while the involvement of the rectum (10%), omentum and peritoneum (5%) is very rare. The GISTs of Meckel's diverticulum are extremely rare,[5] especially if you consider that only 2% of the population has this kind of diverticula confirmed by autopsy studies, with a percentage that varies between 0.4 and 4.5%.[11]

GIST, now recognized as a separate tumour entity defined as mesenchymal tumors that express KIT protein or have an activating mutation in a class III receptor tyrosine kinase gene, the PDGFR-α gene, which encodes the platelet derived growth factor receptor-alpha, a tyrosine kinase protein.[12]

The most frequent symptom is gastrointestinal bleeding related to the size of the tumour, with diameter of 2 cm and ulceration of the gastric-duodenal mucosa, while at the Meckel's diverticulum it is due to ectopic localization of the gastric mucosa.[13] The cause of bleeding is determined by a superficial necrosis of the mucosa and relative ulceration for the development of GIST at the sub mucosa which determines compression of the surface layers.[14] The growth of the tumour is progressive, circumscribed, well encapsulated,[15] sometimes the symptoms are of secondary liver metastases and they are diagnosed with casual CT, MR or during surgical treatment, for the primary lesion.

The GISTs of duodenum and stomach presenting with gastrointestinal bleeding are diagnosed at endoscopy, while lesions in the small intestine are identified by conventional radiology of digestive tract, or better by CT, MR or PET, the last one for the recognition of distant metastases.[15] The limits of endoscopy are due to the sub mucosal location of tumor and the high risk of bleeding which may be effectively controlled by endoscopic guidance. Endoscopy with biopsy is only a preliminary step to surgical treatment.[16]

The selective angiography allows a localization of bleeding and the simultaneous embolization of the vessels that causes hemorrhagic damage.[17] The angiography is mainly indicated in GIST which originates from duodenum, jejunum and Meckel's diverticulum because the embolization reduces the vascularity, minimizing the bleeding risk. The preoperative angiography in a case of tumour of the duodenum reported, allowed the embolization of the pancreatico-duodenalarch, improvement of anaemia and reducing the size of GIST with a safe subsequent surgical treatment.

The FNAB provides a histological diagnostic framework for the research of the encapsulated margins of neoplastic mass, for an adequate adjuvant therapy: it must be carried out with echo or radio-guided technique.[15,18]

Surgery is considered the standard treatment for non-metastatic GIST with en bloc resection and clear margins. There is little evidence to support local/regional lymphadenectomy as GISTs rarely metastasize to lymph nodes.[7] Laparoscopy is indicated for GIST smaller than 8 cm[15]. The minimally invasive surgery treatment has a diagnostic role for GIST, especially for the rare forms with difficulty in preoperative evaluation, also for gastric cancer of the anterior wall with encapsulated lesion that ensures a complete mechanical resection maintaining the margins of oncological safety.[19] Although the role of technology is still controversial for the presence of any recurrence at the point of insertion of the trocar.

Targeted therapy with imatinib, a KIT tyrosine kinase inhibitor, is considered the standard treatment for metastatic GISTs.[7] Pretreatment with imatinib has been an attractive option for GISTs if *en bloc* resection is impossible because of the bulkiness or anatomical location of the tumor. Fiore et al. reported that all patients with GISTs showed a median tumor size reduction of 34% following preoperative imatinib therapy for a median of 9 months with tolerable toxicities[6]. Importantly, the clinical response to imatinib depends on the mutational status of c-kit and platelet-derived growth factor receptor alpha (PDGFRA). Although controversy still remains regarding the optimal duration of neoadjuvant imatinib therapy, the drug has typically been administered for 6 to 12 months.[20] Since resistance to imatinib has been reported, close monitoring is crucial to achieve the best surgical timing; otherwise the opportunity for surgical excision may be missed.[6] In a case of Meckel's diverticulum GIST, treatment with imatinib mesylate has been reported by Khoury et al., but the impact on the clinical behavior of the disease has not been described.[21]

There are little prognostic data regarding GISTs and current prognostic indicators are based on consensus guidelines. The clinical behavior of a GIST is strongly related to its size and mitotic activity: tumour diameter of greater than 5 cm and a high mitotic count exceeding five mitotic figures per 50 high powered fields on light microscopy are indicative of higher malignant potential. Other suggested factors indicative of poor prognosis include tumour perforation, tumour necrosis, high cellularity and marked pleomorphism.[7] Patients with a small bowel localization do worse than those with stomach GIST as reported by De Matteo et al.[9]

Contrast Enhanced CT is a valuable procedure for detection of local recurrence, distant metastases and for surveillance after surgery. If a localized recurrence is detected, the

patient may be treated with repeated resection to prevent complications and to attempt a cure.

Our literature search revealed a total of 42 case reports of GIST of MD including ours (Table:1). GIST of MD occurs

Table 1: Reported cases of GIST of Meckel's Diverticulum in the literature

S. No	Authors	Age/sex	Presentation	Follow up
1	Calderale sm et al. 1997[22]	69/female	Ovarian cyst	Not reported
2	Johnston et al. 2001[23]	58/male	Right iliac fossa pain. Mimicking appendicitis	Not reported
3	Stolk et al. 2002[12]	50/male	Melena for 5 days, lower GI bleed	Not reported
4	Fruhauf Ch et al 2002	61/-	Perforation	Not reported
5	Lorusso et al. 2003[5]	55/male	Mass on CT	Not reported
6	Szentpali et al. 2004[24]	70/male	Perforation	Not reported
7	Barbulescu et al. 2005[25]	69/male	GI bleed	Not reported
8	Hager et al. 2005[26]	75/male	Perforation	Not reported
9	Mijandrusic Sincic et al. 2005[27]	81/male	Crohn's with perforation	Not reported
10	Biemans et al. 2005[28]	-	GI bleed	Not reported
11	Khoury II et al. 2006[21]	28/male	Acute intestinal obstruction with mass	Treatment with imatinib mesylate (outcome not specified)
12	Morimura Y et al. 2006	Female	Ovarian mass	Not reported
13	Chandramohan et al. 2007[6]	65/male	Acute intestinal obstruction with mass	Uneventful postoperative period
14	Komen et al. 2007[29]	79/male	Rectal bleeding GI bleed and mass on CT	Not reported
15	Macaigne et al. 2007[30]	66/female	Rectal bleeding GI bleed	Not reported
16	De la Morena et al. 2007[31]	47/female	ovarian mass	No evidence of disease five years after surgery
17	Kosmidis C et al. 2009[32]	69/male	Synchronous colorectal adenocarcinoma and GIST in the Meckel's Diverticulum -an accidental discovery on laparotomy	Not reported
18	A Hakeem et al. 2008	46/male	GI bleed	Not reported
19	Wennervalt et al. 2009[33]	51/-	GI bleed	Not reported
20	Richard Woolf et al. 2009[34]	59/male	Acute right iliac fossa pain, localised peritonitis perforation	No evidence of disease one year after surgery
21	Michele Sciapi et 2009[35]	68/male	Perforation	Not reported
22	Leung U et al. 2010[36]		Acute intestinal obstruction	Not reported
23	Van Loo et al. 2010[13]		GI bleed	Not reported
24	Dogrul et al. 2010[37]	86/female	Perforation	Not reported
25	Yueh-Hung Chou, MD 2011[38]	76/female	Perforation	Not reported
26	Caricato M et al. 2010[39]	-	Perforation	Not reported
27	WK Mitchell et al. 2011[40]	39/male	Mass on CT	No evidence of disease two years after surgery
28	Mitura et al. 2012[41]	63/female	Perforation	Not reported
29	Fevzi CENGIZ et al. 2012[42]	53/female	Volvulus and acute intestinal obstruction	No evidence of disease two years after surgery
30	Selim Sozen MD et al. 2012[43]	62/female	Perforation	Not reported
31	Lopez et al. 2013[44]	-	Perforation	Not reported
32	Koppad et al. 2013[45]	-	Not reported	Not reported
33	Jigar Vipulshah et al. 2013[46]	30/female	Tubo ovarian mass	Not reported
34	Jigar Vipulshah et al. 2013[46]	50/male	Acute intestinal obstruction	Not reported
35	Goyal R et al. 2013	23/male	Perforation	Not reported
36	Amendolara M et al. 2014[47]	51/female	Ovarian cyst	No evidence of disease one year after surgery
37	Jarrar MS et al. 2014[48]	52/female	Pelvic mass	Not reported
38	Ikemura M et al. 2015[49]	82/male	Perforation	Not reported
39	Omerza CR et al. 2016[50]	-	-	Not reported
40	Englert-Golon M et al. 2016[51]	71/female	Synchronous endometrial adenocarcinoma and GIST in Meckel's diverticulum- accidental discovery on laparotomy	Not reported
41	Hemanth V et al. 2016	53/female	Ovarian mass	Not reported
42	Hemanth V et al. 2016	45/female	Ovarian mass	Not reported

more commonly in the older population group (60% are>50 years old) with the median age being 60 years. These are 1.2 times more common in males. Small bowel GISTs have a range of presenting features, including abdominal pain, an abdominal mass, gastrointestinal bleeding, small bowel obstruction, weight loss, fever, abscess or perforation.

Perforation is the most common presentation of a Meckel's GIST (13 case reports).Gastro intestinal bleeding occured more frequently owing to the increased vascularity of the tumor (8 case reports). Bowel obstruction with a mass was reported in 5 cases, whereas a bowel mass identified on imaging with no obstruction was reported in 2 cases. Another interesting presentation of a Meckel's GIST is that mimicking an acute appendicitis in a 58 year old male.[34]

Englert-Golon M[51] reported a case of synchronous colorectal adenocarcinoma with Meckel's GIST and Kosmidis C[32] reported a synchronous endometrial adenocarcinoma with Meckel's GIST. Amendolara et al[47] reported 6 cases of rare locations of GIST, two in the duodenum, two in omentum and peritoneum, one in the rectum and one in the Meckel's Diverticulum. Pérez-García R et al[52] reported two cases of small bowel GIST mimicking ovarian masses, one with a persisting Meckel's Diverticulum, not included in this series.

Meckel's GIST presenting as a pelvic mass mimicking an ovarian pathology has been reported in the literature (6 case reports). The median age for this group is a decade earlier than the general group, giving them a high chance of being diagnosed as ovarian masses rather than GIST. A preoperative diagnosis of an ovarian mass with a surprising preoperative Meckel's GIST would need a change in the plan of management, especially requiring bowel resection and anastomosis. A bowel preparation would give a better outcome, had it been suspected preoperatively. The anatomical location of the Meckel's diverticulum in the terminal ileum and huge tumors like GIST involving it has a higher chance of occupying the pelvis by effect of gravity and mimicking an ovarian mass. We had the opportunity of adding two such case reports to the literature.

CONCLUSION

The GIST localization to the duodenum, omentum, peritoneum, rectum are rare and of the Meckel's diverticulum is an exceptionally rare one as Meckel's Diverticulum is located in only 2% of population.

Meckel's diverticular tumours, though rare, may mimic other pathologies of the abdomen, therefore should be taken into consideration during the differential diagnosis of abdominal and pelvic tumours, especially in cases where imaging modalities doesn't pinpoint a definitive diagnosis. We present these case reports due to the rarity of presentation of a Meckel's Diverticulum GIST mimicking an ovarian mass. Perforation of the tumor being the most common presentation, the role of prophylactic Meckel's diverticulectomy requires further evaluation.

CONSENT FOR PUBLISHING

Informed consent has been taken from both the patients for publishing the images.

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