

Meckel Diverticulum Harboring a Rare Gastrointestinal Stromal Tumor

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Background: Tumors within a Meckel diverticulum are a rare complication observed in only 0.5%-3.2% of symptomatic cases. The majority of tumors are benign, but some malignant tumors, such as gastrointestinal stromal tumors (GISTs) can occur.

Case Report: We report the case of a 48-year-old female who presented with severe abdominal pain and nausea and was found to have a GIST arising from a Meckel diverticulum.

Conclusion: The differential diagnosis of a pelvic mass in a middle-aged female presenting with gastrointestinal symptoms must remain broad. With an atypical presentation site, distinguishing benign tumors from malignant tumors such as GISTs is of paramount importance.

Keywords: *Gastrointestinal stromal tumors, intestine–small, Meckel diverticulum*

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INTRODUCTION

Meckel diverticulum results from incomplete closure of the vitellointestinal duct and affects 2% of the population, representing the most common congenital anomaly of the small intestine.¹ Most cases are asymptomatic, but when a Meckel diverticulum is symptomatic, adults can present with bleeding, obstruction, diverticulitis, or perforation, with the majority incidentally diagnosed during laparotomy or laparoscopy.¹ Tumors within a Meckel diverticulum are a rare complication observed in only 0.5%-3.2% of symptomatic cases.¹ The majority of tumors are benign, but some malignant tumors, such as gastrointestinal stromal tumors (GISTs) can occur.

CASE REPORT

A 48-year-old white female presented with severe abdominal pain and nausea for the previous 24 hours, having experienced intermittent mild abdominal pain for the past 6 months. She presented with symptoms of small bowel obstruction. A longitudinal transabdominal ultrasound demonstrated a normal fluid-distended bladder and right adnexal mass measuring 6.3 × 4.4 cm and separate from the uterus (Figure 1, arrow). Vascular flow of the adnexal mass was seen on Doppler imaging with spectral analysis. Axial abdominal computed tomography (CT) images with oral and intravenous (IV) contrast displayed a right lower quadrant heterogenous mass (Figure 2, straight arrow) within a loop of ileum with contrast and air along its border (Figure 2, curved arrow). Coronal CT with oral and IV contrast displayed a right adnexal mass (Figure 3, arrow). A

small bowel mass with partial surrounding air and oral contrast was seen on sagittal CT with oral and IV contrast (Figure 4, curved arrow).

The patient underwent robotic hysterectomy and in the operating room was found to have a pelvic mass measuring 8.5 cm in greatest dimension adhered to the right pelvic side wall and attached to the small bowel and some omentum. The surgeon performed laparoscopic small bowel resection with primary anastomosis. Pathologic and subsequent immunohistochemical (IHC) analyses were performed. The sample involved the muscularis propria and submucosa of

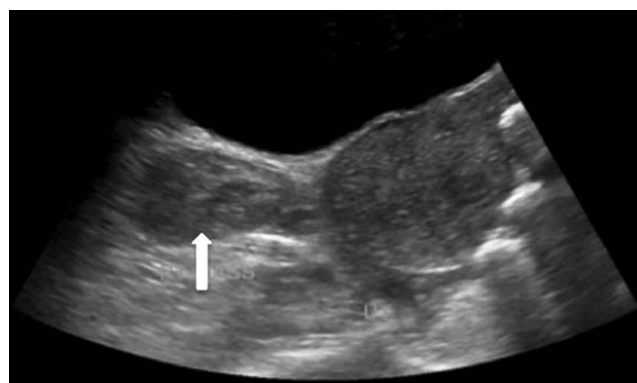


Figure 1. Longitudinal transabdominal ultrasound demonstrates a normal fluid-distended bladder, the uterus, and a 6.3 × 4.4-cm right adnexal mass (arrow) separate from the uterus.



Figure 2. Axial abdominal computed tomography images with oral and intravenous contrast demonstrate a right lower quadrant heterogenous mass (straight arrow) within a loop of ileum with contrast and air along its border (curved arrow).

the small bowel with focal necrosis. IHC staining for CD117, a smooth muscle actin, was positive. CD34, desmin, and S100 were negative (Figure 5).

Imaging and IHC analysis confirmed the patient had a Meckel diverticulum harboring a GIST. One month later in

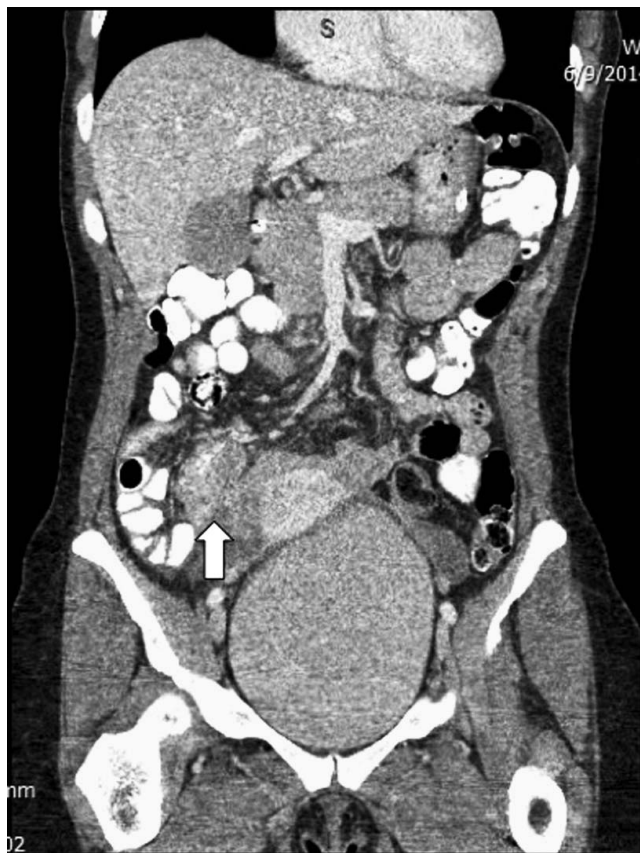


Figure 3. Coronal computed tomography with oral and intravenous contrast displays a right adnexal mass (arrow).



Figure 4. Sagittal computed tomography with oral and intravenous contrast displays a small bowel mass with partial surrounding air and oral contrast (curved arrow).

the clinic, she was administered imatinib but had a severe allergic reaction to the drug. The allergic reaction was treated with cetirizine for 1 week, and the patient was switched to prednisone when she came back to the clinic later that week. However, she developed adverse side effects and is now on only patient-determined supplements (iron, mushrooms, antioxidants, and vitamin D) and following up every 3 months with a pelvic and chest CT with routine blood work. She remains symptom-free and is seeking other opinions.

DISCUSSION

Tumors within a Meckel diverticulum are a rare complication. The majority include benign tumors such as leiomyomas, lipomas, and angiomas. Although less common, malignant neoplasms typically include carcinoid tumors (44%), mesenchymal tumors (35%), and adenocar-

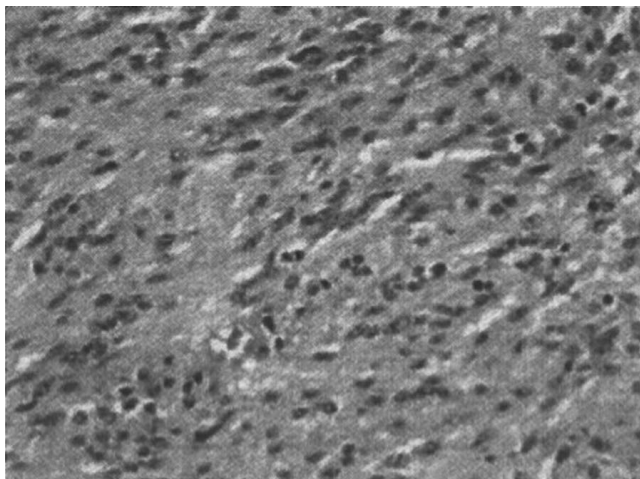


Figure 5. Immunohistochemical staining was positive for CD117. CD34, desmin, and S100 were negative. The tumor involves the muscularis propria and submucosa of the small bowel with focal necrosis.

cinomas (16%).^{2,3} Sixty to 70% of GISTs have been reported to arise in the stomach; 20%-30% in the small intestine; and <10% in the esophagus, colon, and rectum.⁴ Uncommon extraintestinal sites of GISTs are the omentum, mesentery, and retroperitoneum. However, GISTs arising from a Meckel diverticulum are extremely uncommon, with only a handful of known cases reported to date.¹⁻⁵

CONCLUSION

The differential diagnosis of a pelvic mass in a middle-aged female presenting with gastrointestinal symptoms must remain broad. With an atypical presentation site, distinguishing benign tumors from malignant tumors such as GISTs is of paramount importance.

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REFERENCES

1. Yahchouchy EK, Marano AF, Etienne JC, Fingerhut AL. Meckel's diverticulum. *J Am Coll Surg*. 2001 May;192(5):658-662.
2. Cates JM, Williams TL, Suriawinata AA. Intraductal papillary mucinous adenoma that arises from pancreatic heterotopia within a meckel diverticulum. *Arch Pathol Lab Med*. 2005 Mar; 129(3):e67-e69.
3. Carpenter SS, Grillis ME. Meckel's diverticulitis secondary to carcinoid tumor: an unusual presentation of the acute abdomen in an adult. *Curr Surg*. 2003 May-Jun;60(3):301-303.
4. Sozen S, Tuna Ö. A rare case of perforated Meckel's diverticulum presenting as a gastrointestinal stromal tumor. *Arch Iran Med*. 2012 May;15(5):325-327. doi: 012155/AIM.0016.
5. Hager M, Maier H, Eberwein M, et al. Perforated Meckel's diverticulum presenting as a gastrointestinal stromal tumor: a case report. *J Gastrointest Surg*. 2005 Jul-Aug;9(6):809-811.
6. Clinical vignettes/case reports – biliary/pancreas. *Am J Gastroenterol*. 2015 Oct;110 Suppl 1:S40-S550. doi: 10.1038/ajg.2015.270.

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