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Gastrointestinal duplication cyst with ectopic gastric mucosa detected on ^{99m}Tc-pertechnetate SPECT-CT scintigraphy: a case report

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ABSTRACT

Background: Gastrointestinal (GI) tract duplication, or GI duplication cyst, is a rare congenital abnormality that may arise anywhere from the GI tract. We present a unique case of a 2-year-old girl having a large GI duplication cyst with ectopic gastric mucosa (EGM) diagnosed on ^{99m}Tc pertechnetate single-photon emission computed tomography-computed tomography (SPECT-CT) scintigraphy.

Case Presentation: A 2-year-old girl with a history of fresh bleeding per rectum and abdominal distention was referred to the nuclear medicine department for Meckel's scanning to rule out Meckel's diverticulum. Meckel's scanning along with SPECT-CT showed abnormal radiotracer accumulation in the dilated gut loops in the right side of the abdomen simultaneously with that of the stomach, indicative of EGM in GI duplications cyst.

Conclusion: We conclude that although GI duplication cyst with EGM is a rare entity, it should be kept in mind while interpreting the Meckel's scan. ^{99m}Tc pertechnetate SPECT-CT scintigraphy is a useful modality though not commonly used for the preoperative diagnosis of GI duplications cyst with EGM. This case report emphasizes the use of ^{99m}Tc pertechnetate SPECT-CT scintigraphy while evaluating the patients with lower GI bleed.

Keywords: Ectopic gastric mucosa, gastrointestinal duplication cyst, Meckel, scintigraphy, SPECT-CT.

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Introduction

Gastrointestinal (GI) tract duplication or GI duplication cyst is a rare congenital anomaly of the digestive tract with unclear pathogenesis. Clinical presentation and imaging findings are highly variable depending on the size, location, type, mucosal pattern, and presence of complications. Ectopic gastric mucosa (EGM), another rare condition, is usually present in the small intestine and may be associated with GI duplication cyst [1]. Abdominal scintigraphy with ^{99m}Tc pertechnetate single-photon emission computed tomography-computed tomography (SPECT-CT) scintigraphy may be useful in the preoperative diagnosis since the radiotracer is only accumulated by functioning gastric mucosa.

Case Description

A 2-year-old girl presented with a history of fresh bleeding per rectum off and on for 1 year. She was anemic and had a history of multiple blood transfusions for the past 1 year. Her hemoglobin was 6.0 g/dL (12-16 g/dL), and total leukocyte count was 6,800/µL (4,500-11,000/µL) with normal coagulation parameters. Her stool examination was positive for occult blood. She was referred to the nuclear medicine department, for ^{99m}Tc Meckel scan. Her abdominal ultrasonography (USG) showed single fluid distended prominent bowel loop with the largest luminal diameter of 35 mm in the right upper abdomen (Figure 1).

^{99m}Tc pertechnetate scintigraphy revealed abnormal radiotracer uptake in the right side of the abdomen which increases in intensity simultaneously with that of the stomach. To further characterize this abnormal uptake in the right side of the abdomen, ^{99m}Tc pertechnetate SPECT-CT scintigraphy was done, and SPECT-CT images characterized this abnormal uptake as uptake in dilated gut loops (Figure 2).

Since the ^{99m}Tc pertechnetate is accumulated in the functioning gastric mucosa, ^{99m}Tc uptake in the dilated gut loops on ^{99m}Tc pertechnetate SPECT-CT scintigraphy made the diagnosis of GI duplication cyst with EGM.

Discussion

GI duplication cyst is an uncommon congenital malformation of the GI tract. It may be single or multiple and may occur anywhere along the digestive tract. The small intestine

is most commonly involved accounting for about 50% of all the lesions [2]. It affects 1 in every 4,500 individuals, as estimated by autopsy reports, and predominantly occurs during childhood [3]. Most of the GI cysts do not present with debilitating symptoms. However, the symptoms may include abdominal distention, vomiting, bleeding, and a palpable abdominal mass, with complications such as perforation, intussusception, bowel obstruction, volvulus, and associated malignancy depending on the size and the presence or absence of complicating hemorrhage, infection, rupture, or respiratory tract involvement [4].

Ectopic gastric mucosa (EGM) in the GI tract though rare is one of the differentials among the other causes of lower GI bleeding in children such as polyps, clotting disorders, arteriovenous malformations, and Crohn's disease [5]. The presence of gastric mucosa within duplication cysts has been reported to be 17%-36% [6]. The length of the involvement by EGM usually ranges from a few to several centimeters. The diagnosis is usually made during laparotomy for the complications of EGM, such as perforation or obstruction. ^{99m}Tc pertechnetate scintigraphy is commonly used for the diagnosis of Meckel's diverticulum, but it is less commonly used for the pre-operative diagnosis of such cases.

The usual imaging modalities employed to make the diagnosis of GI duplications are USG and CT, but, to ascertain the presence of EGM, a ^{99m}Tc pertechnetate scan is used. ^{99m}Tc pertechnetate is accumulated by EGM in a duplication cyst, similar to that of uptake in gastric mucosa within a Meckel's diverticulum, and this phenomenon may be useful to diagnose this condition. The use

Figure 1. *USG showed single fluid distended prominent bowel loop with a maximal luminal diameter of 35 mm in the right upper abdomen.*

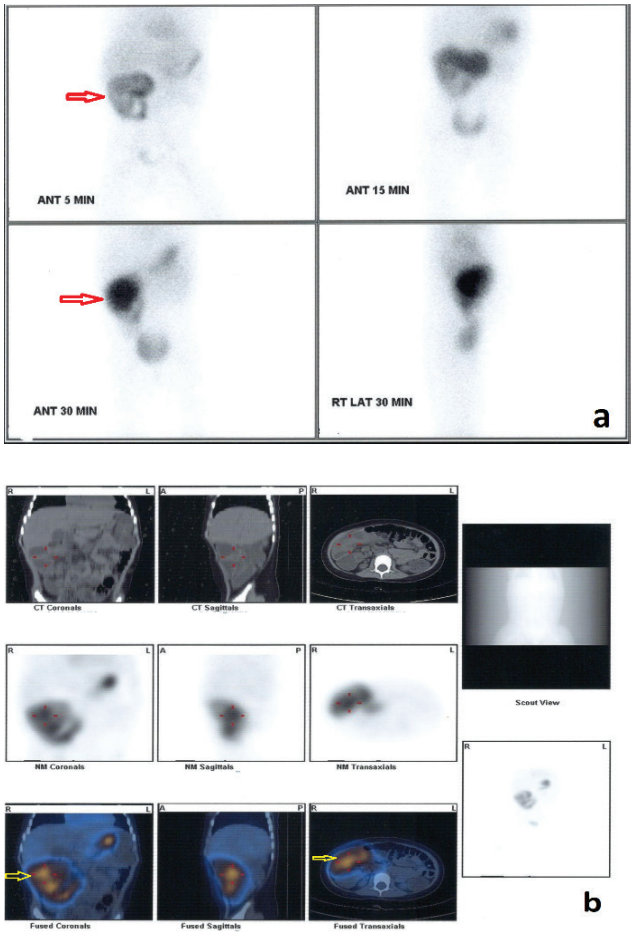

Figure 2. *(a) ^{99m}Tc pertechnetate scintigraphy showing increased tracer concentration in the right side of the abdomen on planer imaging (red arrows) and (b, c, and d) corresponding activity in dilated gut loops as seen in SPECT-CT images (yellow arrows).The intensity is seen to increase simultaneously with that of the stomach indicative of ectopic functioning gastric.*

of concomitant SPECT-CT imaging further improves the diagnosis by accurate localization.

The sensitivity of ^{99m}Tc pertechnetate imaging for the detection of GI duplication cyst is reported to be 75% [7]. False-positive uptake of this radiotracer may be seen in intussusception, focal hyperemia or bowel inflammation, GI bleeding unrelated to EGM, uterine blush, retention of the tracer in the urinary collecting system, and vascular lesions such as hemangiomas and arteriovenous malformations [4]. However, the concomitant use of SPECT-CT may play an important role in making the final diagnosis and excluding the false-positive causes. The additional effective dose delivered from the CT component of SPECT-CT was estimated to be 3.8 mSv [8]. Hence, the need of doing CT should be judged keeping in mind the risk–benefit ratio and should be done only in those cases where justified and add some additional benefit to solve the suspicion.

The previous studies showed that the uptake pattern in EGM is highly variable and tends to decrease during the course of imaging due to dilution with intestinal secretions. The locally accumulated tracer also gets washed away by peristaltic activity [7,9]. However, in the present case, the ^{99m}Tc is retained in the right side of the abdomen and increased in intensity simultaneously with that of the stomach, and the pre-operative diagnosis of GI duplication cyst with EGM was made by using ^{99m}Tc pertechnetate SPECT-CT scintigraphy that confirms the uptake in dilated gut loops.

Conclusion

We conclude that any abnormal uptake on Meckel's scan should be thoroughly evaluated using ^{99m}Tc pertechnetate SPECT-CT scintigraphy, and clinicians should keep in mind such rare entities such as GI duplication with EGM while interpreting the scan. Furthermore, it emphasizes the use of ^{99m}Tc pertechnetate SPECT-CT scintigraphy while evaluating the patients with lower GI bleed.

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List of Abbreviations

EGM	Ectopic gastric mucosa
GI	Gastrointestinal
SPECT-CT	Single-photon emission computed tomography-computed tomography

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Funding

None.

Consent for publication

Written informed consent was obtained from the parents of the patient to publish this case in a medical journal.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

Authors' contribution

FG and MBI were responsible for recognizing the importance of publicizing the role of nuclear medicine in the diagnosis of this rare pathology and lessons associated with this case to the wider medical community. FG, MBI, and OQ diagnosed the patient and contributed in writing the draft of the case report. MS and MN contributed to writing the discussion part of the case report. All the authors approved the final version of the manuscript.

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