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*Case report*

# SMALL INTESTINE INTUSSUSCEPTION DUE TO GASTROINTESTINAL STROMAL TUMOUR IN PREGNANCY: A CASE REPORT

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**Abstract**

Gastrointestinal stromal tumour (GIST) is very rare in pregnancy and only a few cases have been described in the literature. We present a case of a 38-year-old primigravida, presented with non-specific symptoms for the first time in the second trimester. Due to the non-specificity of the symptoms on one hand and the rarity of the tumour on the other, it took a long time for the final diagnosis to be made.

Accidentally, on a routine obstetric ultrasound examination, a solid tumour formation was observed, localized under the lower pole of the left kidney. On MRI of the abdomen, in front of the left kidney there was a tubular structure, in close relation with small intestine, suspected for intussusception. At 28 weeks of gestation, an exploratory laparotomy was performed with resection of the involved part of the jejunum and TT anastomosis. Pregnancy was terminated electively, by caesarean section, in 38+6 gestational weeks.

The clinical presentation of the GIST depends on the primary location of the tumour. Due to the extremely rare occurrence of these tumours in pregnancy, there is no solid scientific evidence for the most appropriate time of their treatment and the time of termination of pregnancy. The biggest challenge in pregnancy is timely diagnosis and treatment, without impact on the foetus. A multidisciplinary approach is needed. In our case, the severity of the mother's symptoms outweighed the danger to the foetus from general anaesthesia and surgery itself.

**Keywords**: GIST, pregnancy, non-specific symptoms, multidisciplinary

## Introduction

Gastrointestinal stromal tumour (GIST) is the most common mesenchymal (non-epithelial) neoplasm of the GI tract. Nevertheless, GISTs are rare neoplasms and represent only 1-2% of all primary GI neoplasms. They are very rare in pregnancy and only a few cases have been described in the literature[4]. Therefore, their diagnosis and treatment in pregnancy is a challenge for obstetricians and surgeons. We present a rare case of a mother with GIST and non-specific symptoms diagnosed in the second trimester of pregnancy.

**Case report**

A 38-year-old woman, gravida-1 para-0, visited an internal medicine clinic at 18 weeks gestation for the first time due to persistent vomiting and great weight loss in the last month. There were no significant medical conditions before. Laboratory examinations supported a newly diagnosed hyperthyroidism, there was also elevated hepatic transaminase activity, mild hyperbilirubinemia and anaemia.

Therapy with propylthiouracil was initiated, with concomitant regulation of thyroid status, as well as a gradual decline in transaminases activity and clinical improvement of the patient. Therefore, she was discharged from the clinic.

After two weeks she visited the University Clinic for Obstetrics and Gynaecology due to worsening of the condition, persistent vomiting, epigastric and groin pain and progressive weight loss. She was hospitalized at the Department for urgent gynaecologic conditions under the diagnosis of severe hyperemesis in pregnancy. In the meantime, a diagnostic amniocentesis was performed due to the triple X syndrome result of a non-invasive prenatal screening test for aneuploidy. During hospitalization she was treated symptomatically with occasional improvement of the condition and periods of deterioration. The patient had a significant weight loss and BMI of 17.7. At the 24th gestational week she was transferred to the obstetric intensive care unit and during a routine ultrasound control of the foetus, a tumour formation was detected, localized under the lower pole of the left kidney with dimensions of about 80x60 mm. On control ultrasound by an internist, a pronounced stagnation in the stomach and duodenum was found, probably due to a distal obstruction, and a structure measuring 80x60 mm with good central vascularization, suspected of intussusception. On MRI of the abdomen, in front of the left kidney there was a tubular structure with diameter of 65 mm, in close relation with small intestine, in favour of intussusception (Figure 1). Tumour markers levels were in reference range.

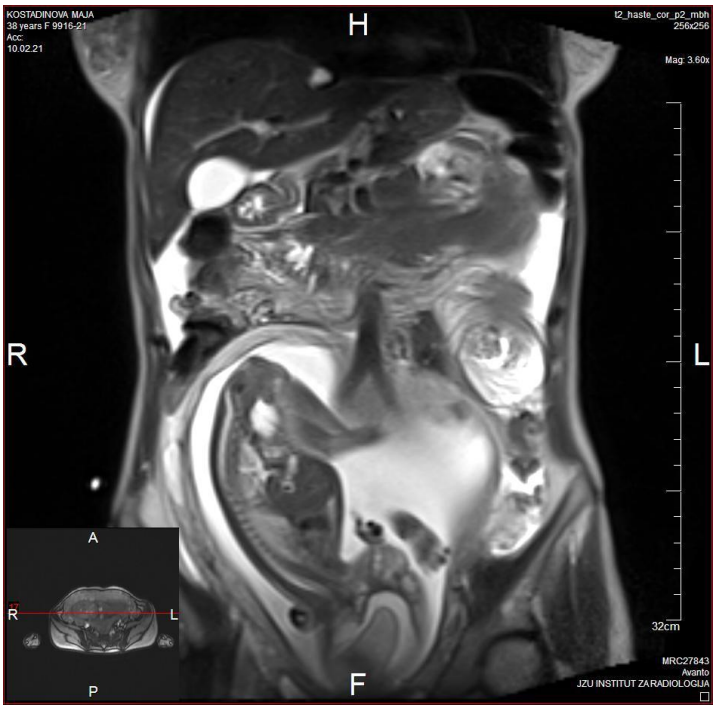


**Fig. 1.** MRI of the abdomen, in front of the left kidney there is a tubular structure with diameter of 65 mm, in close relation with small intestine, in favour of intussusception

Upper gastrointestinal endoscopy revealed stenosis at the level of jejunum with necrosis. However, it was not possible to determine whether the cause of intussusception was a luminal

substrate. Due to the findings of the above examinations as well as the further deterioration of the general condition of the patient, the digestive surgeon together with the obstetrician opted for an exploratory laparotomy at 28 weeks of gestation with a working diagnosis of intussusception of the jejunum. Resection of the involved part of the jejunum was performed with TT anastomosis. After opening the specimen, a polypoid intraluminal lesion on a wide base with diameter of about 4 cm was revealed (Figure2).

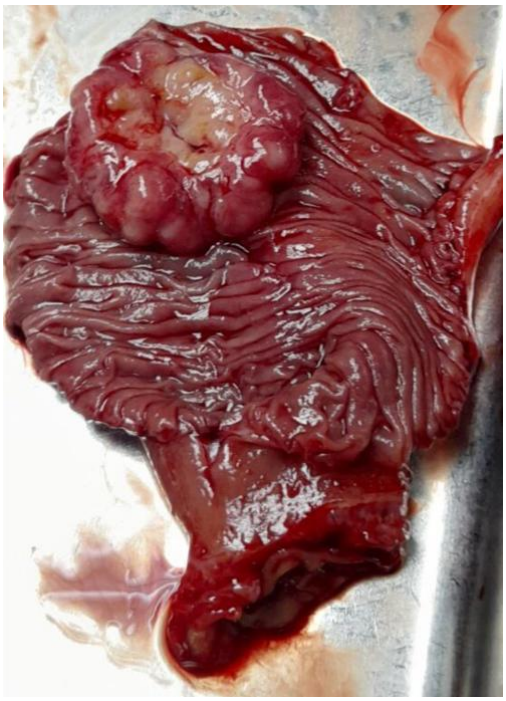

**Fig. 2.** Surgical specimen - a polypoid lesion on a wide base with diameter of about 4 cm

The histological diagnosis was GIST. The resected tumour was 4x4 cm in size, with negative surgical resection margins. Immunohistochemical staining was specific for GIST: CD34 (+) and CD117 (+). Histological findings of 3 mitoses at 50 HDF and size <5 cm are indications of a tumour with a low risk of recurrence.

The postoperative period was uneventful, with continuation of pregnancy at home and regular perinatal examinations every month. Two months after the surgery, BMI was 20.5. She delivered electively, by caesarean section, in 38+6 gestational weeks. The newborn was in a good condition, with a birth weight of 3020 grams, corresponding to the gestational age. The patient and the newborn left the hospital on the third postoperative day in a good general condition.

At a routine check-up by an oncologist 6 months after the surgery, a CT scan of the abdomen was performed and tumour markers were examined. The results were in reference values.

**Discussion**

GIST is a rare neoplasm, accounting for only 1 to 2 percent of primary gastrointestinal cancers, with a median age of diagnosis between 65 and 69 years[2]. Therefore, the occurrence of these neoplasms in pregnancy comes down to a few cases. In the absence of scientific evidence for their management during pregnancy, patients should be treated multidisciplinary by surgeons, gastroenterologists, oncologists and obstetricians.

GISTs are a separate entity from other mesenchymal tumours of the GI tract, and are believed to originate from interstitial cells of Cajal or so-called GI pacemaker cells. They can occur along the entire GI tract, from the oesophagus to the anus, most commonly in the stomach (40-60%) and jejunum/ileum (25-30%)[3].

The clinical presentation depends on the primary location of the tumour[4]. Those originating in the upper GI tract present with dysphagia, bleeding or obstructive jaundice, while those originating in the colon or rectum present with constipation and intestinal obstruction. 10-20% have metastatic disease at the time of diagnosis, with the most common site of metastasis in the liver, omentum or peritoneum. GIST is quite often identified incidentally in asymptomatic patients during procedures for another indications.

In suspected patients, the diagnosis is made by MRI/ CT as well as upper/lower digestive endoscopy. The definitive diagnosis is made by histopathology and immunohistochemistry. CD 117 or the almost universal overexpression of the tyrosine kinase KIT receptor is the most characteristic marker of GIST[2]. All GISTs have a potential to metastasize. There are prognostic models based on known risk factors that determine the relative risk of recurrence and metastasis. The most significant risk factors are tumour size and mitotic index. Primary localization is another independent risk factor, so primary gastric tumours are thought to have a better prognosis than those occurring in the small intestine, colon, rectum or mesentery[3].

Due to the extremely rare occurrence of these tumours in pregnancy, there is no solid scientific evidence for the most appropriate time of their treatment and the time of termination of pregnancy[5]. The biggest challenge in pregnancy is timely diagnosis and treatment, without impact on the foetus. Aggravating factors for early diagnosis in pregnancy are the age of onset, which does not coincide with the usual age of onset of these tumours in the literature (65-69 years of age) and diverts the clinician from the differential diagnosis, as well as the non-specific symptoms that are often attributed to the pregnancy itself[6].

The primary treatment for localized disease is surgery, and the best chance of cure is when complete surgical resection of the tumour with clean margins is achieved[7]. A multidisciplinary approach is needed. In our case, the severity of the mother's symptoms outweighed the danger to the foetus from general anaesthesia and surgery itself.

The literature also describes cases of elective termination of pregnancy between 35 and 37 weeks of gestation and postponement of surgical treatment postpartum[3]. The management of these tumours in pregnancy is almost identical to that outside of pregnancy, and pregnancy very rarely affects the prognosis.

In patients at high risk of recurrence, adjuvant therapy with imatinib-targeted therapy with tyrosine kinase inhibitors is postoperatively recommended. The safety of this therapy during pregnancy has not yet been established[6].

Cases of foetal malformations, miscarriages, and elective termination of pregnancy during imatinib therapy have been reported[6]. Further studies are needed to establish recommendations for GIST treatment during pregnancy. Until then, all decisions are made individually, with the help of a multidisciplinary approach.

**Conclusion**

Gastrointestinal stromal tumours are extremely rare in pregnancy. All decisions regarding the diagnosis and treatment of these tumours in pregnancy should be made in a multidisciplinary manner, taking into account the modest experience so far with these neoplasms in pregnancy described in the literature. It is also very important to know the prognostic factors and modalities of treatment when making decisions. In our case, the patient received a better treatment and outcome thanks to the multidisciplinary approach.

*Conflict of interest statement*. None declared.

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