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Ectopic Decidua Presenting With a Sigmoid Bowel Perforation: A Case Report

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Abstract

The occurrence of ectopic decidua has been observed mostly in the ovaries, uterus and cervix. Ectopic gastrointestinal deciduosis is a physiological condition. Only several cases of ectopic decidua, causing complications have been presented in literature. To the best of our knowledge, sigmoid or any other bowel perforation, caused by deciduosis, has not been previously reported in literature.

Case presentation: A 40-year old patient primigravida (32 weeks of gestation) was admitted with a severe abdominal pain and obstipation. An abdominal sonography was performed but no visual cause of the pain was diagnosed. Two days after admission the episode of severe pain reappeared. An abdominal sonography and x-ray showed "free air" under both domes of diaphragm. A laparotomy was performed. No perforation of hollow viscus was found. On the fifth postoperative day, the patient started to complain about acute abdominal pain. The decision was made to perform relaparotomy (a Caesarean section and reexploration of abdominal cavity during the same operation). During the relaparotomy a part of sigmoid colon was visually altered. A microperforation on tumour-like structures was found in the sigmoid colon. A Sigmoid resection was performed. Currently the patient has no residual complaints and is enjoying her motherhood.

Conclusion: We assert that ectopic decidua can result in a perforation of the hollow viscus, which is a life-threatening condition and should always be taken into account seriously. We believe that sigmoid microperforation could have found, we had taken a more aggressive approach and performed a Caesarean section during the first operation.

Keywords: Deciduosis; Complications; Sigmoid colon perforation

Abbreviations: ICU: Intensive Care Unit

Introduction

The first case of ectopic (extrauterine) decidua was described by Walker [1]. The occurrence of ectopic decidua has been observed mostly in the ovaries, uterus and its cervix. Usually ectopic extrauterine deciduosis appear during pregnancy. However, cervical and ovarian decidual lesions may appear in non-pregnant patients, mostly in relation to gestagen therapy or even without it. A peritoneal localization is less frequent and usually is an asymptomatic incidental finding [2,3]. Ectopic gastrointestinal deciduosis is a physiological condition hypothetically resulting from progesterone-induced metaplasia of the subserosal cells [4]. Only several cases of ectopic decidua causing complications, such as intra-abdominal bleeding [5] or obstruction of labour [6], severe abdominal pain similar to the pain of appendicitis or mechanical bowel obstruction [7] were presented in literature. To the best of our knowledge, sigmoid or any other bowel perforation has not been previously reported. Tumour-like and mass forming intraperitoneal ectopic deciduosis of the pregnant patient with deep invasion to sigmoid colon causing perforation with favorable outcome is presented in this case report.

Case Presentation

A 40-year-old primigravida patient (32 weeks of gestation) was admitted to the Department of Obstetrics with severe abdominal pain on the right side of the abdomen and obstipations; the patient did not have nausea or vomiting. The patient upon the admission was consulted by an abdominal surgeon and urologist. An abdominal sonography was performed but no visual cause of the pain was observed. Laboratory tests showed elevated white blood cells count (WBC – 12×10^9/l) and C-reactive protein (CRP – 54.06 mg/l). After administering intravenous fluids and analgetics her condition improved.

Two days after the admission the episode of severe pain reappeared. The second abdominal sonography was performed. "Free air" under both domes of diaphragm was found. Abdominal X-ray confirmed the diagnosis.

The patient was transferred to the Department of Abdominal Surgery and was operated upon. An upper-middle laparotomy was performed since perforated duodenal ulcer was suspected. As no perforation could be found, intraoperative upper endoscopy and water-air test were performed (both negative). Without being able to determine the cause of "free air" in the abdominal cavity, the operation was terminated. The drain was left in subhepatic space.

After the operation the patient was treated in the ICU. During the

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following four days the patient`s condition did not have any negative dynamics. She was treated with Ceftriaxon 2 g 2 times per day, Metronidazole 500 mg 3 times per day, intravenous fluid infusions, anticoagulants (Fraxiparine 0.2 mg daily). CRP`s level normalized, as well as WBC count and no fever was observed.

On the fifth postoperative day our patient started to complain about acute abdominal pain again. It was localized in the left lower part of the abdomen. Laboratory findings showed increased CRP (209 mg/l) and WBC (2325×10^9/l) count. The decision was made to perform a Caesarean section and re-exploration of abdominal cavity.

A baby girl was born. Apgar score was 8 at 1 minute and 10 at 10 minutes after the delivery. During the relaparotomy, small amount of fibrin threads were observed in the minor pelvis. Approximately 20-25 centimeters of sigmoid colon were altered: the intestinal wall was thickened and covered with soft tumour-like tissue. No bowel perforation was observed. Intraoperative cold section biopsy was taken. The result was "myxoid-like tissue with smooth muscle miocytes". There was not enough data to confirm a tumour. Saline solution was poured into the abdominal cavity and air was pumped through the anus. Eventually bubbles were observed from a microperforation in one of the tumour-like structures. Because the changes in the sigmoid were tumour-like, without having been able to deny the possibility of malignancy, a sigmoid resection was performed. The descending colon was anastomosed with the rectum, the abdominal cavity was drained.

The postoperative period was smooth, without any complications. The patient was followed-up for fifteen months after the second operation. No complications were observed at the time. Currently the patient has no residual complains, she is breastfeeding and enjoying her motherhood.

Pathology

Macroscopy: Dark, red, soft nodules in the submucosa, muscular is propria, subserosa of the resected large bowel. Identical exophytic nodules in subserosal space of the small bowel and uterus (biopsies) (Figure 1).

Methods: All specimens were fixed in 10% buffered formalin, embedded in paraffin. 4 μm sections were stained with haematoxylin-eosin (3 μm sections used for immunohistochemistry). Immunostaining was performed with antibodies for Pan Cytokeratin Dako; dilution 1:100; EDTA Ag retrieval), Vimentin (Dako; dilution 1:800; EDTA Ag retrieval), Ki67 (Dako; dilution1:200; EDTA Ag retrieval), ER/ PR (Ventana; ready for use; Ag retrieval solution CC1), Calretinin (Dako; dilution 1:30; EDTA Ag retrieval), Placental alkaline phosphatase (Dako; dilution 1:40; EDTA Ag retrieval), Cytokeratin 5 (DAKO; dilution 1:100; EDTA Ag retrieval).

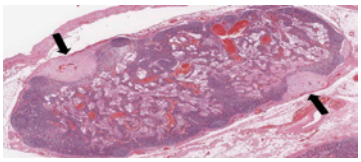

Figure 1: Resected large bowel with polypoid dark red colored exophytic masses in the subserosal space.

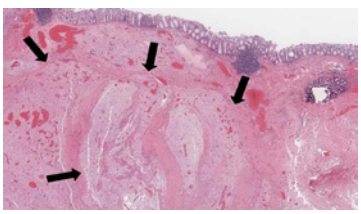

Figure 2: HE 1.5x. Large bowel's wall with ectopic decidual nodules in the subserosa, muscularis propria and submucosa (arrows).

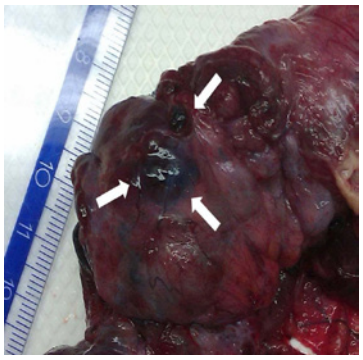

Figure 3: HE 2x. Ectopic decidual nodules in the sinuses of the lymph node (arrows).

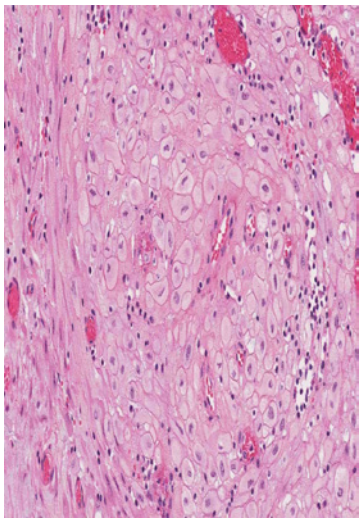

Figure 4: HE 20x. Decidual nodule in the large bowel's wall: The large cells with glassy eosinophilic cytoplasm and distinct borders within capillarised stroma.

Histology: The sigmoid colon showed the ectopic decidua confluent nodules in subserosa, muscularis propria and submucosa, composed of large polygonal cells with glassy eosinophilic cytoplasm with distinct borders (Figure 2,4). Identical nodules were found within sinuses in the pericolonic lymph nodes (Figure 3), uterine and small intestine's subserosal space (biopsies were taken from these parts too). The decidual foci showed typical immunoprofile: progestine receptors (PR) /Vimentin diffusely positive, estrogen receptors (ER) focally positive. Ki67 proliferative index was very low < 1%. Pancytokeratin, Calretinin, Placental alkaline phosphatase and Cytokeratine 5 stains were negative.

Discussion

Ectopic deciduosis is a known phenomenon, described in different locations as cervix uteri [8], ovary [9], fallopian tubes, appendix [10], peritoneum [6], omentum [4], diaphragm, liver, spleen, pleura [11], lymph nodes [12], renal pelvis [13], skin [14].

Clinically extra uterine ectopic deciduosis may easily simulate disseminated tuberculosis [3] or metastatic disease [2].

In some rare cases (such as ours) extrauterine deciduosis may simulate primary neoplastic growth, forming large polypoid nodules usually in subserosal space or even in the deep tissues of visceral organs.

The main differential diagnosis of ectopic tumour-like deciduosis should be with rare malignant deciduoid mesothelioma, which arises commonly in the peritoneum of young women [15]. In our case the nodules of deciduosis, despite infiltrative and deep growth pattern, spreads serosal surface differently, than mesothelioma. The immunoprofile of deciduosis cells was different: CK5 and Calretinin negative and estrogen/progesterone receptors positive. Low Ki67 proliferative index, rare mitoses serve as additional argument against mesothelioma diagnosis. The macroscopy of the deciduosis lesions (hemorrhagic dark red nodules) was different from white and firm nodules/plagues in mesothelioma cases. Metastatic carcinomas are usually pan cytokeratin positive and display more atypical cytological features. It is interesting that in rare cases deciduosis masses may be cytokeratin positive, too. Therefore, our findings were in line with immunoprofile described in decidual cells [16].

The deep seated deciduosis masses may complicate the labour [6], cause serious complications, such as appendiceal perforation, acute bowel obstruction [17], intraperitoneal hemorrhage [19], peritonitis, adhesions. Even fatal outcome was observed [19].

In our case, due to pregnancy and enlarged uterus, there was no possibility to visualize and explore the suppositional perforation area in the minor pelvis during the first operation.

Although there are opinions that deciduosis does not need any therapeutic interventions or operations and it regresses postpartum without any complications [7], in our case this does not apply, because the patient could have died without an appropriate operation.

Conclusion

In our case the wall of sigmoid colon was deeply infiltrated by deciduosis nodules and became fragile. We can speculate that mechanical pressure from pregnant uterus together with intracolonic gas pressure caused sigmoid rupture with the clinical course described above.

We believe that sigmoid microperforation could have been found had we taken a more aggressive approach and performed a Cesarean section during the first operation. Various ways to find the perforation were used but none was successful. However, speculating about these reasons has little meaning. We hereby state that ectopic decidua can result in a perforation, which is a life-threatening condition and should always be taken into account seriously.

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