CASE REPORT

Case Report: A unique case of a retroperitoneal abscess during pregnancy complicated with an ileo-sigmoid-vesical fistula in a patient with active Crohn's disease [version 1; peer review: 1 approved with reservations, 1 not approved]

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Abstract

The enterovesical fistula is an uncommon condition resulting from complications of inflammatory diseases such as Crohn's disease. While erosion of an abscess can result in the formation of enterovesical fistula, early diagnosis of abscesses is crucial in order to avoid complications. This case is a retrospective analysis of the unusual combination of events that contributed to the misdiagnosis, further complications in a pregnant 28-year-old female diagnosed with Crohn's disease. The patient presented a retroperitoneal abscess, which went undiagnosed and complicated with an ileo-sigmoid-vesical fistula during pregnancy. She complained of right sciatica, cruralgia, and a complete inability to lift her right leg as well as recurrent urinary infections, which were treated by multiple courses of antibiotics. The chronic situation led to the restricted growth of the fetus, which made the fetus not tolerate the labor. Emergency C-section was done due to fetal distress, but baby did well and did not need NICU. None of the gynecological examinations, ultrasound, X-ray, CT-scan, cystography, and colonoscopy performed before, during and after pregnancy were able to explain her symptoms. Fecaluria is the symptom that ultimately directed toward the presence of an eventual fistula five months after delivery. The MRI diagnosed the retroperitoneal abscess and an ileo-sigmoid-vesical fistula, which were treated by total parenteral nutrition followed by ileo-colo resection. An ileostomy was maintained for four months before anastomosis. Sciatic pain and psoasitis in a context of Crohn's disease should alert physicians to the possible presence of a retroperitoneal abscess, which should be monitored by MRI. In addition, recurrent urinary infections during pregnancy in a context of Crohn's disease can be sign of a formed enterovesical fistula. The multiple antibiotic courses, while allowing the continuity of the pregnancy, also contributed to further delay of

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Approval Status

	1	2
version 1		
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1. Siv Fonnes , University of Copenhagen, Herlev, Denmark 2. Rahul Hegde , Yale New Haven Health, Bridgeport, USA		
Any reports and responses or comments on the article can be found at the end of the article.		

diagnosis by decreasing inflammation.

Keywords

case report, sciatic pain, recurrent urinary infection, abscess, appendicitis, fistula.

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Introduction

Enterovesical fistulae are uncommon conditions that are most commonly diagnosed based on clinical evidence. While only about 2% of patients with Crohn's disease develop enterovesical fistulae, ileovesical fistulae are the most common type^{1,2} and the ileo-sigmoid-vesical fistula remains an exceptional case. One of the mechanisms of the formation of an enterovesical fistula the erosion of an abscess³⁻⁵. The importance of early diagnosis of such abscess, in order to avoid complications, is illustrated in this case. A retrospective analysis of the case allows us to propose important symptoms that shall alert on the presence of an abscess and/or an enterovesical fistula. In addition, we will comment on the benefit and inconvenient of using prophylactic antibiotic courses in this case.

Case report

Patient information and clinical findings

The patient was a 28-year-old female, with no past surgical intervention and no known family history of inflammatory bowel disease. She was diagnosed in France with an inflammation of the last 10 cm of the ileum leading to the diagnosis of a Crohn's disease and was treated with an initial dose of 50 mg/day of prednisolone and 50 mg/day of azathioprine for 8 weeks. As her symptoms started to fade, prednisolone dose was progressively decreased to 5 mg/day.

At first visit, the primary concern of the patient was the continuous sciatic pain and an inability of her to lift her leg, with no fever. She reported having had an episode a high fever and kidney pain while on holiday in Tunisia two weeks earlier. Recurrent urinary infections and abdominal stiffness were retrieved at her monthly examinations during her pregnancy follow-up and with elevated C-reactive protein levels of 225 mg/l (normal range <5 mg/l). Because of the pregnancy, limited imaging options were available, and ultrasounds could not give a clear diagnosis. Instead, appendicitis and kidney stones were proposed several times. After pneumaturia and fecaluria appeared, MRI was performed and finally revealed the presence of a retroperitoneal abscess and an ileo-sigmoid-vesical fistula.

Timeline of diagnostic and therapeutic interventions

The time elapsed between the first complaint and the resolution of the underlying cause of the symptoms was 18 months. This patient was diagnosed by several specialties and in different countries as the case happened during the relocation of the patient to a new country. For these reasons, this timeline is based on reported facts from the patient and from the images, lab analyses and reports she could provide. The case happened in 2012, and this patient has been followed up for eight years.

February 2011: Patient complains from high fever, abdominal pain and is diagnosed by ultrasound with a right kidney dilation. She is given paracetamol 500 mg every 6 hours and 500 mg oral ciprofloxacin every 12 hours for 7 days.

March 2011: Patient complains for the first time of sciatic pain, cruralgia and inability to lift right leg, while having no fever. She is taking 5 mg/day prednisolone and 50 mg/day

azathioprine for Crohn's disease management. CT-scan for L3–L4 is normal, ultrasound is normal, gynecological examination is normal, hip X-ray is normal. Abdominal CT-scan shows dilation of right ureter and stenosis against the terminal ileum. She is prescribed 500 mg paracetamol every 6 hours.

April 2011: Patient moves to Qatar.

May 2011: Patient continues complaining from sciatic pain and flexing incapacity, when pregnancy is discovered. The patient decided by herself to discontinue azathioprine at this point, but was followed up with a family medical doctor to maintain doses of prednisolone at 5 mg/day. At first emergency hospital admission, patient presents with severe abdominal pain, with right lower abdominal stiffness and a C-reactive protein levels showing an inflammation of the ileum region leading to the diagnosis of appendicitis. Appendectomy is proposed but the patient refuses surgery.

July 2011: Patient is admitted to hospital with severe kidney pain, general fatigue and paleness. She was taking 500 mg paracetamol every 6 hours. Urine had an abnormal brown and cloudy urine color, which lead to the diagnosis of a urinary infection and beginning of pyelonephritis, confirmed by ultrasound. Augmentin (amoxicillin 250 mg and clavulanic acid 125 mg) is prescribed with a dose of 1g every 12 hours for 7 days.

November 2011: The patient is admitted for pre-term labor. Patient is under the same antibiotic and dose as in July 2011 for the recurrent urinary infections and does not present symptoms. Tocolytic medication is given to stop contractions.

January 2012: The patient is admitted for normal labor; however, fetal distress required emergency C-section. The baby weighed 2400 g, but with a good Apgar score and did not need NICU.

February 2012: The patient complains of severe urinary infections and pneumaturia; cystoscopy is normal. She is on auto medication with 500 mg paracetamol every 6 hours. She is again treated with Augmentin (amoxicillin 250mg and clavulanic acid 125mg) at a dose of 1g every 12 hours for 7 days.

May 2012: Colonoscopy is performed but it was not possible to reach the terminal ileum. A few days after the colonoscopy, the patient is admitted to emergency with vomiting, high fever, severe abdominal pain. Ultrasound and CT-scan show inflammation of the right iliac fossa, leading to diagnosis of appendicitis. The patient is convinced of having a fistula. She refuses surgery and is treated Flagyl (metronidazole hydrochloride) 250 mg twice daily for 7 days.

June 2012: The patient reports fecaluria and is admitted for an MRI examination that reveals the presence of a retroperitoneal abscess and an ileo-vesical fistula. She travels to her home country (France), where a new MRI reveals that the fistula is actually an ileo-sigmoid-vesical fistula. She is treated with total parenteral nutrition for 30 days, and with a combination of 500 mg

metronidazole IV and 400 mg ciprofloxacin IV every 12 hours for 4 weeks, after which ileo-colo resection was performed. An ileostomy was maintained for four months before anastomosis.

Follow up

The patient has been followed up for eight years. After resection, an anti-TNF α treatment was proposed but the patient refused and went onto a restrictive diet (the Specific Carbohydrate Diet) a grain-free diet planned to help people with Crohn's disease^{6,7}. The patient has been followed up for eight years, during which she stayed in remission, with normal levels of C-reactive protein, and two pregnancies with babies of normal birth weight. Significant number of adherences resulted from the surgeries happened in 2012 (c-section, ileo-colo resection, and anastomosis surgery), leading to the formation of a peritoneal cyst that reached 13 cm in 2015, and which reduced by itself during the second pregnancy. Adherences led to the complication of the third C-section by a bladder injury of two cm, which healed after one week of urine catheterization. The patient reports having been more sensitive to fungal infections as important candidiasis happened after each of her three pregnancies, requiring the use of a single dose of fluconazole 150 mg and daktarin cream (miconazole 2%).

Discussion

Our retrospective analysis of the events is that a retroperitoneal abscess was formed in February 2011, which resulted in psoasitis, which explains both sciatica and cruralgia. The patient was under prednisolone, which might have increased the risk of infection. Several diagnostic challenges further complicated the case. First, the relocation of the patient led to difficulties to navigate through the healthcare system, as patient visited different specialists (gynecology, urology, internal medicine, gastroenterology, orthopedics). Second, X-ray and CT-scan were not possible during pregnancy, and urinary infection and sciatic pain were attributed to pregnancy. Third, antibiotic courses allowed the patient to enter into a cycle of flare-recovery that delayed understanding of the disease but allowed pregnancy

to pursue to term with no fetal damage. As pregnancy progressed, the gastrointestinal tract was moving in relation to the uterus and bladder, which might have allowed the formation and closure of the fistula at various locations along the urinary tract leading to further confusion.

The pharmacological approach was used to reduce symptoms and allow pregnancy to pursue to term and allowed the patient to continue to work and have a normal active life, principally using paracetamol and antibiotics. However, this contributed to the delay of the diagnosis of the fistula. Nevertheless, as growing evidence on the importance of the gut microbiome during pregnancy are being exposed, it is important to question whether a preferential therapeutic intervention to protect both mother and child is available. Total parenteral nutrition has been shown to successfully resolve enteric fistula in Crohn's disease and a safe option during pregnancy^{8,9}.

To conclude, this case highlights the importance of close monitoring of any symptoms occurring during pregnancy in patients with active Crohn's disease. Sciatic pain and psoasitis in a context of Crohn's disease shall alert on the possible perforation of the ileum and a retroperitoneal abscess, which need to be monitored by MRI¹⁰. Recurrent urinary infections during pregnancy in a context of Crohn's disease can be sign of the presence of an enterovesical fistula. Finally, courses of antibiotics provide a temporary recovery from symptoms, but also a decrease of the inflammation thus masking the lesions observed on imagery at the time of investigation, leading to further delay of correct diagnosis. Investigations shall be performed far from antibiotic courses.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of clinical details was obtained from the patient.

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Current Peer Review Status: ? ✖

Version 1

Reviewer Report 17 August 2020

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Rahul Hegde

Department of Radiology, Yale New Haven Health, Bridgeport, CT, USA

Abstract:

- Although comprehensive, it would be better if the authors would make the abstract concise.

Introduction:

- It would be better to word that 'ileo-sigmoid-vesical fistula is uncommon rather than calling it 'exceptional' which implies that it is very rare, which it is not, especially in Crohn's disease where severe disease can have complex fistulas.

Case report section:

- 'diagnosed by ultrasound with a right kidney dilation'- I am presuming the authors mean right renal hydronephrosis? - Please correct.
- March 2011- What CT was performed? Lumbar spine of abdominopelvic CT? Without or with contrast?
- May 2011- How does CRP lead to the conclusion of inflammation of ileum region? Was imaging performed at this instance?
- May 2012 and June 2012- Unfortunately, this case has no images shown. It is difficult to believe that CT would not pick up a large intra-abdominal abscess.
- Overall, the case is not very novel as complex fistulizing disease is known to happen with Crohn's disease. Multiple shortcomings mentioned above. Lack of figures/imaging is a significant shortcoming.
- I would refer authors to read the following review of fistulizing disease in Crohn's: Booya, F., Akram, S., Fletcher, J.G. *et al.* CT enterography and fistulizing Crohn's disease: clinical benefit and radiographic findings. *Abdom Imaging* **34**, 467–475 (2009).

<https://doi.org/10.1007/s00261-008-9419-1>¹

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Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Partly

Is the case presented with sufficient detail to be useful for other practitioners?

No

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Radiology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to state that I do not consider it to be of an acceptable scientific standard, for reasons outlined above.

Reviewer Report 05 August 2020

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Siv Fonnes

Centre for Perioperative Optimisation, University of Copenhagen, Herlev, Denmark

This is an interesting case report, which underlines that the patient's medical history (in this case Crohn's disease) is of great importance when diagnosing and treating "ordinary" symptoms and complaints (e.g. recurrent urinary infection during pregnancy).

General comments:

Throughout your manuscript, it is not sufficiently clear which of the complaints and findings that covers the patient's complaints and which consist of findings of the clinical examination. Please make this clear throughout the manuscript.

Introduction:

1. The first sentence in the introduction is unclear and has no reference to either of the statements: "Enterovesical fistulae are uncommon conditions that are most commonly diagnosed based on clinical evidence." Please revise it.
2. Your reference 1 concerning the incidence and rate of fistulas for Crohn's patients is a case report, it does not seem plausible that this is the most accurate reference – please consider revising the reference.
3. Please add a verb in the sentence: "One of the mechanisms of the formation of an enterovesical fistula the erosion of an abscess".
4. You write: "One of the mechanisms of the formation of an enterovesical fistula the erosion of an abscess³⁻⁵." Reference 5 is a small case series of 15 patients, consider changing or omitting this reference.

Case report: patient information and clinical findings

1. What year was she diagnosed with Crohn's disease?
2. The time course is a bit hard to follow - when was "the first visit", please specify.
3. Was abdominal stiffness a symptom reported by the patient or a finding of repeated clinical examinations?
4. The sentences "After pneumaturia and fecaluria appeared, MRI was performed and finally revealed the presence of a retroperitoneal abscess and an ileo-sigmoid-vesical fistula" is not accurate. I agree that pneumaturia is a key symptom and should be reported here. But pneumaturia was reported in February 2012, but she was not diagnosed until June 2012. Please revise the sentence.

Case report: Timeline of diagnostic and therapeutic interventions

1. The timeline is a bit hard to follow. Have you considered making a figure along with the description?
2. All the CT scans were they with or without contrast if with contrast was it given i.v., p.o. or rectally? Did any of the CT scans show intravesical air?
3. There are no descriptions of the urine, urinalysis, or analysis of the urine until July 2011. In February and March, there were clinical findings in relation to her right kidney – was there really no information on her urine at these appointments?
4. May 2011: "At first emergency hospital admission, the patient presents with severe abdominal pain, with right lower abdominal stiffness and a C-reactive protein levels showing inflammation of the ileum region leading to the diagnosis of appendicitis." The

sentence should be revised – there are no descriptions of scans that show inflammation of the ileum.

5. In January 2012 the patient had an emergency C-section, was there any information regarding her abdominal cavity in the surgical record?
6. Did the patient only report pneumaturia in February 2012 and not during any of the other visits?
7. How many times did the patient experience recurrent urinary infections, the information is not clear from the timeline?
8. May 2012, “The patient is convinced of having a fistula”, could you please elaborate on how the patient suspected that – and none of the doctors previously?

Discussion:

1. You write: “Second, X-ray and CT-scan were not possible during pregnancy,” – please make it clear how a fistula should be diagnosed e.g. CT with oral or rectal contrast, as the timeline and your discussion do not clearly state that.
2. Please add the importance of the symptoms recurrent urinary infection, pneumaturia, and fecaluria in suspecting the diagnose fistulas.

References:

1. Reference 1 is not cited correctly.
2. Your reference 7 is missing details.

Is the background of the case's history and progression described in sufficient detail?

Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Partly

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Appendicitis, peritonitis, hernia, microbiome, abdominal surgery.

I confirm that I have read this submission and believe that I have an appropriate level of

expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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