# CASE REPORT

**SUMMARY** 

# Abdominal tuberculosis in a spigelian hernia

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Accepted 1 May 2019

Abdominal tuberculosis is a common form of extrapulmonary tuberculosis. Often it is difficult to diagnose due to vague symptoms and lack of clinical findings. Spigelian hernia is a rare type of hernia located in the semilunar fascia of the abdominal rectus muscular sheath. We report on a 19-year-old Greenlandic Inuit man with a spigelian hernia as the primary presentation of abdominal tuberculosis. Abdominal tuberculosis presenting with a spigelian hernia is extremely rare, and the case illustrates that tuberculosis may present in myriad ways. The incidence of tuberculosis in Greenland is among one of the highest in the world, and we also review the history and incidence of tuberculosis in this Arctic country.

## BACKGROUND

Abdominal tuberculosis is a common form of extrapulmonary tuberculosis and compromises about 5%-11% of all cases of tuberculosis.<sup>1 2</sup> The intestines are most commonly involved when abdominal tuberculosis occurs. The peritoneum is seldom affected, and when it is, there is a risk of hernial sacs being involved.

We report on the occurrence of abdominal tuberculosis presenting as a spigelian hernia.

Spigelian hernia is a rare type of hernia located in the fascia semilunaris, which is the caudal most extent of the abdominal rectus muscle sheath.<sup>3 4</sup>

Both abdominal tuberculosis and spigelian hernias are often difficult to diagnose due to vague symptoms and lack of clinical findings. The occurrence of tuberculosis presenting as a spigelian hernia is extremely rare, and we have only found two other cases in the literature.<sup>5 6</sup>

## **CASE PRESENTATION**

A 19-year-old Inuit man from Greenland presented with pain in the upper right quadrant of the abdomen. The pain appeared after he had started a vigorous physical job and was thought to be of muscular origin. He had normal gastrointestinal function. Blood tests showed a haemoglobin of 143.4 (normal 133.7-169.2g/L), leucocytes of 8.7 (normal  $3.5-8.8\times10^{9}/L$ ), normal liver and kidney function tests and a negative Quantiferon test. One year later, he contacted the hospital again as he still suffered from abdominal pain. Clinical examination revealed a palpable mass at the right side of the abdomen, and ultrasound confirmed the diagnosis: a spigelian hernia located in the fascia semilunaris in the abdominal rectus muscle. The patient was scheduled for surgery without further investigations.

#### TREATMENT

An open herniotomy was performed under general anaesthesia. During surgery pus was found at the hernia site. Cultures were taken, and the hernia was repaired. The pus was sent to the laboratory and came back negative for bacterial growth. Unfortunately, no tissue was sent for histopathological examination. The skin incision was closed after cleaning the wound, and a drain was placed. A mesh was not inserted due to the presence of infection. The patient was given perioperative intravenous cefuroxime and metronidazole, and postoperatively the patient was treated with oral ciprofloxacin and metronidazole. After surgery, it turned out that the patient for some time had suffered from tiredness, intermittent fever, and general malaise. He had lost 4 kg in weight.

A laboratory test showed a positive Quantiferon test, which had been negative 1 year ago. Sputum test for mycobacterium was negative. Furthermore a chest X-ray was negative. Blood tests showed a haemoglobin of 140.2 (normal 133.7-169.2 g/L), leucocytes of 10.2 (normal  $3.5-8.8 \times 10^{9}$ /L). Based on the clinical findings (general malaise, abdominal pain, weight loss and intermittent fever), the findings at surgery and the conversion of the Quantiferon test to positive, relevant treatment for tuberculosis was initiated. The patient should have undergone more extensive investigations if the suspicion of abdominal tuberculosis had been raised before surgery.

## **OUTCOME AND FOLLOW-UP**

There were postoperative complications in the form of infection of the skin incision. The wound was opened and left for healing. The patient came to regular check-ups for cleaning and changing of the wound. He received anti-tuberculosis treatment for 9 months with isoniazid, rifampicin, pyrazinamide and ethambutol. His symptoms of abdominal pain, general malaise, weight loss and fever disappeared.

At the 9 months' check-up, there was no evidence of recurrence of tuberculosis and the treatment was terminated.

#### DISCUSSION

A spigelian hernia is a rare type of ventral abdominal hernia, and represents less than 0.12% of all abdominal hernias.<sup>3</sup> Spigelian hernias may cause unspecific symptoms like weight loss and abdominal tenderness. Sometimes sharp pain is present at the hernia site. Diagnosis of a spigelian hernia by physical examination is typically very difficult.<sup>4</sup> The combination of unspecific abdominal symptoms

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**To cite:** Aalling L, Penninga L. *BMJ Case Rep* 2019;**12**:e227638. doi:10.1136/bcr-2018-227638

# Unusual association of diseases/symptoms

and difficulties with physical examination may cause diagnostic delay, which was also the case in our patient.

Abdominal tuberculosis is reported in 5%–11% of extrapulmonary tuberculosis, whereas 65%–78% involves the gastrointestinal tract and often causes ulcers or strictures.<sup>12</sup>Extra-intestinal tuberculosis with involvement of the peritoneum and lymph nodes may occur, but is more unusual. Peritoneal tuberculosis is difficult to diagnose due to very non-specific symptoms, which include fever, abdominal pain, anorexia and weight loss.<sup>7</sup>

Only two other cases of abdominal tuberculosis in a spigelian hernia have been reported.<sup>5</sup> <sup>6</sup> In the first case, the patient presented with abdominal pain for 10 days, and symptoms of intestinal obstruction. Surgery was performed, and an ileal loop was found in a spigelian hernia as well as a peritoneal fluid collection. Perioperative tissue was examined, and PCR for mycobacterium tuberculosis was positive.<sup>5</sup> In the second case, a tuberculous ovary was found in a spigelian hernia.<sup>6</sup>

Definitive diagnosis of abdominal tuberculosis can be challenging as it requires identification of mycobacterium tuberculosis bacilli in the peritoneum or ascites or histopathological confirmation. Acid fast staining is normally negative and cultures are only positive in 30%–40% of cases.<sup>5</sup> This makes bacteriological confirmation of tuberculosis difficult. PCR is a good method to establish the diagnosis. In our patient, no definitive diagnosis could be made, though abdominal tuberculosis was very likely based on the clinical findings (general malaise, abdominal pain, weight loss and intermittent fever), the findings at surgery, the conversion of the Quantiferon test to positive and the symptoms disappeared during anti-tuberculosis treatment. Furthermore, tuberculosis is endemic in Greenland and the patient was not vaccinated.

Cases of abdominal tuberculosis presented with other types of abdominal hernias have been reported.<sup>8-14</sup> In these cases, the tuberculosis presented as a thickening and nodulation of the hernia sac and in some with the presence of excessive peritoneal fluid. In all of these cases, the patient presented symptomatic with a hernia.

Our patient lives in Arctic Greenland. In the 1950s, the population of Greenland had the highest rate of tuberculosis worldwide.<sup>15</sup> Hence, a nationwide neonatal vaccination programme with the bacille Calmette-Guerin (BCG) vaccination was introduced in 1955. In 1989, this intervention was phased out as tuberculosis was nearly absent from Greenland, and the vaccination programme ended in 1991.<sup>15</sup> From 1991 to 1997, the incidence rate of tuberculosis was doubled and the vaccination programme was reinitiated.<sup>16</sup> Children born between 1991 and 1997 were offered vaccination after reinitiation of the vaccination programme, but many were not reached or did not accept the vaccination. Due to the lack of vaccination, there is a higher incidence of tuberculosis in people born between 1991 and 1997 compared with the rest of the population, and men are more often affected than women.<sup>16</sup> Our patient was born between 1991 and 1997, and had not received BCG vaccination.

Although the incidence of tuberculosis in Greenland is declining due to better surveillance, management and infection control, tuberculosis is still endemic.<sup>16</sup> It is important to investigate for tuberculosis when patients present with unspecific symptoms, given that tuberculosis continues to present itself in myriad ways. What should have been a simple herniotomy for a spigelian hernia, turned out to be a rare presentation of abdominal tuberculosis.

## Learning points

- Both a spigelian hernia and abdominal tuberculosis are difficult to diagnose due to vague symptoms and lack of clinical findings.
- Abdominal tuberculosis in hernias often presents with thickening and nodulation of the hernia sac—but not always.
- Greenlandic patients born between 1991 and 1997 are at higher risk of tuberculosis if not bacille Calmette-Guerin vaccinated.

**Contributors** LA and LP had the idea. LA extracted the data and prepared the first draft. LP critically revised the draft, and both authors accepted the final version of the manuscript.

**Funding** The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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