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Case report

Adnexal torsion caused by tuberculous pyosalpinx: An uncommon case report of urogenital tuberculosis

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ABSTRACT

Background: According to the World Health Organization, TB is a global public health problem and it remains in 2020 the deadliest infectious disease in the world, ahead of Covid19 (*Global Tuberculosis Report 2020*, 2020). Morocco is an endemic area with more than 30,000 new cases of all forms of tuberculosis each year (*Plan stratégique national 2018-2021 de lutte antituberculeuse*, 2018). UGTB is the second most frequent localization after lymph node involvement and is responsible for 30 to 40% of all extrapulmonary cases.

Case presentation: We hereby present the uncommon case of a 27-year-old virgin woman with unremarkable medical and surgical histories, who presented at the emergency department for left-sided pelvic pain of acute installation. An exploratory laparotomy using a Pfannenstiel incision, demonstrated a peritoneal effusion of low abundance with a twisted left ovary and a huge pyosalpinx. Detorsion was then carefully performed, with improvement in color of the ovary and decrease in edema within 10 min. Histopathological study of the Fallopian tube biopsy revealed granulomatous abscessificated salpingitis with genital tuberculosis. Thus, the patient received her anti-tuberculosis treatment with a course of 6 months of 2HRZE/4HR.

Conclusions: In view of the resurgence of cases of tuberculosis of all forms, the importance of prevention and screening should not be underestimated, especially in endemic areas. In fact, only BCG at birth and the correct treatment of any primary tuberculosis infection, whatever its location, will make it possible to reduce the consequences of this affection and avoid the tragedy of the home without children.

1. Introduction and importance

According to the World Health Organization, TB is a global public health problem and it remains in 2020 the deadliest infectious disease in the world, ahead of Covid19 [1]. Morocco is an endemic area with more than 30,000 new cases of all forms of tuberculosis each year [2]. UGTB is the second most frequent localization after lymph node involvement and is responsible for 30 to 40% of all extrapulmonary cases [3–5]. UGTB is characterized by insidious onset and nonspecific symptomatology often resulting in delayed diagnosis and rapid progression [3,4]. We will review the current knowledge of this common disease through the presentation of a singular case report.

2. Case presentation

We hereby present the uncommon case of a 27-year-old virgin woman with unremarkable medical and surgical histories, who presented at the emergency department for left-sided pelvic pain of acute installation, evolving for 5 h, associated with vomiting. She however denied association with night sweats, fever or weight loss. In terms of family history, the patient's father had pulmonary tuberculosis 17 years back, for which he was treated for 6 months. On physical examination, the patient was conscious with a GCS of 15, VAS of 10, afebrile with hemodynamic and respiratory terms as follows: Blood pressure at 110/60 mm Hg, heart rate at 103 bpm, respiratory rate at 15 cycles/min and oxygen saturation of 99%. Abdominal examination found a soft abdomen with the presence of an increased sensitivity of the left iliac

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fossa. Speculum examination did not demonstrate any vaginal discharge and bimanual pelvic examination demonstrated cervical excitation with significant discomfort in the left adnexa. Transabdominal and transvaginal pelvic ultrasound showed a 95 × 43 mm dilated tubular structure within the left adnexa that has no color flow on Doppler suggesting a large twisted left ovary. Blood tests revealed CRP of 120 mg/L and a WCC of 12.5 × 10^9/L.

On the basis of these findings, the patient immediately underwent an exploratory laparotomy using a Pfannenstiel incision, demonstrating a peritoneal effusion of low abundance. After adhesiolysis, the uterus was seen grossly normal looking together with the right Fallopian tube and ovary. The left ovary was however in severe ischemia, twisted with the distal two third of the left fallopian tube 2 times around themselves, with the presence of a huge pyosalpinx involving its entire length (Fig. 1). Detorsion was then carefully performed, with improvement in color of the ovary and decrease in edema within 10 min. After salpingostomy, massive pus poured out of the tube and samples for culture were taken. Peritoneal toileting was done with eight litters of warm normal saline. Then, a temporary abdominal drain was inserted. The postoperative course was unremarkable and the pain resolved completely after surgery. The patient was discharged on the 5th post-operative day in stable condition.

Histopathological study of the fallopian tube biopsy revealed granulomatous abscessificated salpingitis with suspected genital tuberculosis due to the presence of several epitheloid and gigantocellular granulomas centred by a caseous necrosis. The abscess sample taken during the laparotomy (Fig. 2) was initially Ziehl-Neelsen stain negative but the culture in the Löwenstein-Jensen medium was found positive for *Mycobacterium tuberculosis*. The search for associated pulmonary tuberculosis by smear microscopy for AFB three days in a row and a pulmonary CT-scan was unsuccessful. Thus, the patient received her anti-

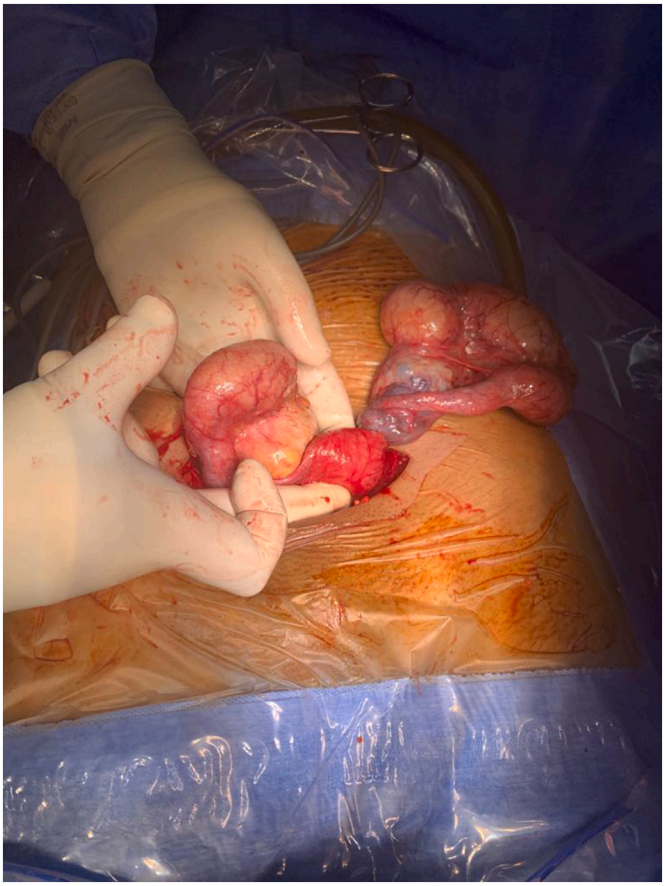

Fig. 1. Adnexal torsion caused by huge pyosalpinx.

Fig. 2. Abscess sample taken during the laparotomy.

tuberculosis treatment with a course of 6 months of 2HRZE/4HR (2 months of the isoniazid, rifampin, pyrazinamide and ethambutol association followed by 4 months of isoniazid and ethambutol biotherapy). Despite difficulties of therapeutic compliance, the patient completed her treatment and she was declared cured of her genital tuberculosis.

3. Clinical discussion

3.1. Urogenital tuberculosis

According to the World Health Organization, TB remains in 2020 the world's most deadly infectious disease; it claims more than a million lives each year and affects millions more [1]. About a quarter of the world's population is infected with *Mycobacterium tuberculosis* (rarely *Mycobacterium bovis* and/or atypical mycobacteria) and the proportion of EPTB accounts for 10% of TB cases [1,3,4]. UGTB is the second most frequent localization after lymph node involvement and is responsible for 30 to 40% of all extrapulmonary cases [3–5].

UGTB is most often the result of blood or lymphatic transmission from latent primary infection primarily from the lungs or other organs, but sometimes directly from adjacent organs (intestine or lymph node) [3,4]. It can rarely be sexually transmitted through semen infected with active genitourinary tuberculosis in the male partner [6,7]. Our patient was probably infected by her father, then she may have developed a latent primary infection for years which then spread to the genital tract, the sexual route being eliminated because of her virginity. The forms of female genital tuberculosis during the period of genital activity represent almost all cases and are observed between 20 and 35 years old, but like our patient, many of these cases are the consequence of a primary infection that dates back to post puberty.

The initially paucisymptomatic and nonspecific presentation as well

as the insidious evolution of UGTB are at the origin of an often-important diagnostic delay [8]. The most frequent clinical presentations are ureteral strictures for urinary forms, epididymal nodule in men and chronic salpingitis in women for genital forms like our case [8,9]. The definitive diagnosis of TB is only made by bacteriological examinations. However, urogenital tuberculous lesions, in particular the genital forms of women, are very frequently paucibacillary and bacteriological examinations may not reveal the bacterial strain. In our case, abscess sample's direct examination using the Ziehl-Neelsen stain was negative and it was only thanks to the culture on the Löwenstein-Jensen medium that we were able to highlight the responsible agent and thus initiate the treatment.

3.2. Adnexal torsion

Torsions of the adnexa are a common pathology, since they represent 2.5 to 7.4% of women consulting for acute pelvic pain according to Huchon et al. [10], and 3% of gynecological emergencies according to Houri et al. [11]. Adnexal torsion is achieved by a rotation of at least one turn of the adnexa on the axis defined by the lumbo-ovarian ligament and the tuboovarian ligament, involving the fallopian tube and the ovary as in our case, or the ovary alone and less frequently the fallopian tube alone [12].

The clinical results of ovarian torsion vary depending on how the rotation is set up. With progressive torsion, lymphatic drainage is compromised first, leading to an increase in ovarian size due to lymphatic edema [13–15]. It is followed by venous obstruction and hemorrhagic infarction [13,15,16]. The final stage is the interruption of the arterial blood supply which may be inaugural in the event of sudden torsion. If left untreated, this ischemia will lead to necrosis of the ovary, fallopian tube or entire appendix and may decrease subsequent fertility in young patients [17]. It can also be complicated by gangrene, infection, peritonitis and in rare cases death (mainly in young girls) [18,19].

Torsions of the adnexa most often occur in women of childbearing age and are usually indicative of an ovarian abnormality [10,11]. The main risk factors for adnexal torsion are as follows: pregnancy, induction of ovulation, increase in the size of the ovary (in particular by benign tumors) [10–12]. Moreover, as suggested by Huchon et al. [12], the right side is implicated in two thirds of cases of adnexal torsion, which can be explained by a right utero-ovarian ligament physiologically longer than the left and/or by the presence of the sigmoid on the left decreasing the space necessary for a torsion. The presence of paratubal cysts, weighing down the tube alone, may be responsible for isolated torsion of the tube or of the entire adnexa [12]. Likewise, the presence of hematosalpinx, hydrosalpinx or pregnancy is a risk factor for isolated tube torsion [12]. In our case precisely, the presence of a large pyosalpinx led to the torsion of the adnexa.

The suspicion of adnexal torsion implies emergency surgery but as the literature shows the preoperative diagnosis is difficult with according to White et al. [20], a median duration of symptoms before diagnosis of 3 days and a diagnostic delay of 22 h. Moreover, according to the study by Huchon et al. [10] on the effectiveness of the preoperative diagnosis, it is only correct in 23 to 66% of cases, explained by a great disparity in the modes of presentation. Huchon et al. [21] developed an aid score for the preoperative diagnosis of adnexal torsions based on five criteria: unilateral lumbar or abdominal pain (25 points), pain duration less than 8 h at first presentation (25 points), vomiting (20 points), absence of leucorrhoea and metrorrhagia (20 points), and finally ovarian cyst larger than 5 cm by ultrasonography (10 points). Values below 35 for this score would make it possible to rule out the diagnosis of adnexal torsion with a risk of less than 5%, while a score greater than 60 would find the presence of an adnexal torsion in 70% of cases, involving the performance of an emergency laparoscopy. For patients with a score between 35 and 60 and therefore an intermediate risk, the authors suggested additional explorations that could be carried out: in particular the diffusion MRI described by Fujii et al. [22] having a specificity greater than 90% for the diagnosis of adnexal torsion or even studies on interleukin 6 intake by Daponte et al. [23]. This score requires large-scale prospective evaluations to be validated but appear promising. Our patient had a score of 75% which would therefore require urgent surgical exploration.

3.3. Management and prognosis

The treatment of UGTB is essentially medical for 6 months divided into intensive quadruple therapy (isoniazid, rifampicin, Ethambutol, pyrazinamide) for two months, then maintenance treatment for 4 months with daily dual therapy (isoniazid, rifampicin). Surgery is indicated in the event of a compressive or fistulized mass [24]. The prognosis of pelvic tuberculosis is linked to infertility in young women and the risk of tuboovarian infertility is estimated at 39%.

4. Conclusion

In view of the resurgence of cases of tuberculosis of all forms, the importance of prevention and screening should not be underestimated, especially in endemic areas. In fact, only BCG at birth and the correct treatment of any primary tuberculosis infection, whatever its location, will make it possible to reduce the consequences of this affection and avoid the tragedy of the home without children [25].

This work has been reported in line with the SCARE 2020 criteria [26].

Abbreviations

TB	Tuberculosis
UGTB	Urogenital tuberculosis
GCS	Glasgow Coma Scale
VAS	Visual Analog Scale
CRP	C-reactive protein
WCC	White cell count
AFB	Acid-fast bacilli
CT-scan	Computerized tomography scan

Availability of data and materials

Supporting material is available if further analysis is needed.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethics approval

Ethics approval has been obtained to proceed with the current study. Written informed consent was obtained from the patient for participation in this publication.

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Aziz Slaoui: study concept and design, data collection, data analysis and interpretation, writing the paper
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Najia Zeraidi: study concept, data collection, data analysis, writing the paper
Amina Lakhdar: study design, data collection, data interpretation, writing the paper
Aicha Kharbach: study design, data collection, data interpretation, writing the paper
Aziz Baydada: study concept, data collection, data analysis, writing the paper.

Declaration of competing interest

The authors declare that they have no competing interests.

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