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The Pseudomyxoma Peritonei in Ouagadougou: Report of Two Cases and Review of the Literature

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Abstract

Introduction: Pseudomyxoma peritonei is a rare pathology requiring, according to international recommendations, multidisciplinary management in a specialized center. The aim of our work was to assess the difficulties of this care in our context through the analysis of two cases managed in Ouagadougou. **Observations:** In the first observation, this was a 60-year-old, male patient, who was diagnosed with a pseudomyxoma peritonei grade I according to the WHO two years after the onset of symptoms. He benefited from cytoreductive surgery and two sessions of adjuvant chemotherapy. Due to lack of sufficient financial resources, he had to interrupt his chemotherapy and died in 2023. For the second observation, it was a 51-year-old, female patient, where the diagnosis of pseudomyxoma peritonei secondary to ovarian and appendicular mucinous cystadeno-carcinoma was retained five months after the onset of symptoms. She underwent cytoreductive surgery with oophorectomy, appendectomy, hysterectomy removing the left ovary and adjuvant chemotherapy courses. The development has been favorable. **Conclusion:** Pseudomyxoma peritonei remains a very rare pathology in our context, often with diagnostic delay and treatment difficulties. They could be somewhat attenuated by establishing a framework for reflection and management of this type of pathology adapted to our realities.

Keywords

Africa, Ascites, Ouagadougou, Burkina Faso, Pseudomyxoma Peritonei

1. Introduction

Pseudomyxoma peritonei is an anatomo-clinical entity characterized by a localized or diffuse accumulation of mucinous materiel in the peritoneal cavity governed by redistribution phenomena, and secondary to a mucinous neoplasia located in the vast majority of cases in the appendix, much more rarely in the urachus, pancreas and ovary [1]. It is a rare pathology whose incidence is estimated at 0.2 per 100,000 people per year worldwide [2]-[4].

Few studies have focused on this pathology in Africa. The first published cases date from 1996 by Zeraidi *et al.* in Morocco [5] and Nguema-Mve and Menye in Gabon [6].

In Burkina Faso, Zida *et al.* [7] reported the first case of pseudomyxoma peritonei in 2010.

Its rarity, lack of awareness and knowledge often lead to delays and misdiagnosis [8]. In France, an organization called the national network for the management of rare tumors of the peritoneum (RENAPE), which includes pseudomyxoma peritonei, exists and allows patients to be referred to expert and skilled centers for appropriate treatment [8].

In sub-Saharan Africa and particularly in Burkina Faso, to our knowledge, no specialized center is available for the management of this condition.

The aim of our work was to evaluate the difficulties of treating pseudomyxoma peritonei in our context through two observations collected in Ouagadougou.

2. Observation 1

A 60-year-old, male patient, residing in Ouagadougou, was seen for a hepato-gastroenterology consultation in August 2018 for abdominal distention associated with weight loss.

The onset of symptoms dates back to August 2017 when the presence of ascites was noted during surgery for right inguinal hernia in a local health center. An abdominopelvic ultrasound performed in September 2017 concluded that ascites was of medium to high abundance, with an infiltrated mesentery, the site of a vesicular image, motivating further exploration by a computed tomography (CT) scan to avoid ignoring pseudomyxoma peritonei. Thoraco-abdominal CT performed in October of the same year revealed the presence of minimal bilateral pleural effusion, abundant ascites, homogeneous hepatomegaly and non-obstructive right renal microlithiasis. The ascites puncture brought back a yellow liquid. Biochemical analysis of the liquid found proteins level at 66 g/l and a cell density estimated at 700/mm^3 including 95% lymphocytes.

The hemoglobin level was 11.8 g/dl. The rest of the biological assessment in-

cluding serum creatinine, blood ionogram, total and conjugated bilirubins, transaminases, fasting blood glucose was normal. An exploratory laparoscopy was proposed but the patient was lost to follow-up. Faced with the persistence of ascites associated with unquantified weight loss, asthenia without associated fever, the patient consulted a faith-based health center from where he was referred to a pulmonology service. There he benefited from anti-tuberculosis treatment for six months without clinical improvement. Given that the condition of the patient was not improving, he presented himself for a hepato-gastroenterology consultation at the Tengandogo University Hospital in August 2018.

The physical examination revealed stage II general condition according to the World Health Organization (WHO) classification; ascites of great abundance.

The paraclinical assessment carried out found:

- Low in proteins level in ascites fluid (<40 g/L), leukocytes (16/mm^3) and absence of pathogenic micro-organisms
- GeneXpert for Mycobacterium Tuberculosis was MTB Negative/RIF;
- Abdomino-pelvic ultrasound fund abundant ascites with diffuse infiltration of mesenteric fat and in particular, a heterogeneous right para-umbilical mass containing mesenteric fat;
- Complementary abdomino-pelvic CT scan found abundant ascites without visible tissue expansive process, particularly in the para-umbilical area;
- Antero-posterior frontal chest X-ray revealed an elevation of the right hemidiaphragmatic dome with bands of atelectasis on the side, requiring additional hepatic assessment;
- Anti HBc Ab were positive, but hepatitis B DNA was undetectable normal prothrombin level of 100%;
- Upper digestive endoscopy, grade I esophageal varices, small volume hiatal hernia, congestive and purpuric corporeal gastropathy and sessile polyps in the antrum and duodenum.

A laparotomy was decided at a Multidisciplinary Consultation Meeting (RCP). In December 2018, an appendectomy was performed with also a removal of gelatinous mass (**Figure 1**), mass sent to the pathological anatomy laboratory.

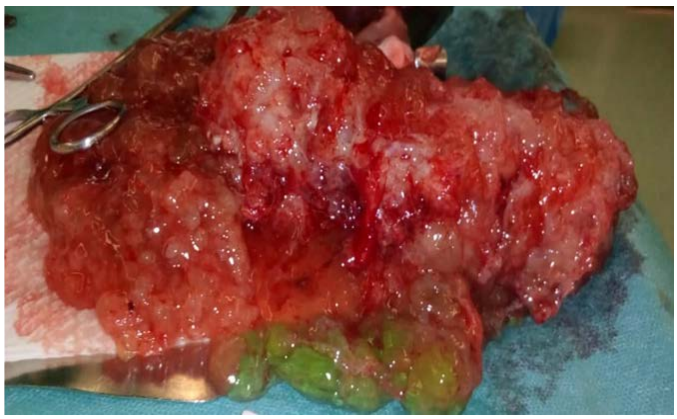

Figure 1. Surgical specimen: gelatinous mass (Dr Sissoko L).

The histological examination concluded to pseudomyxoma peritonei WHO grade I (2010 WHO grading) (Figure 2).

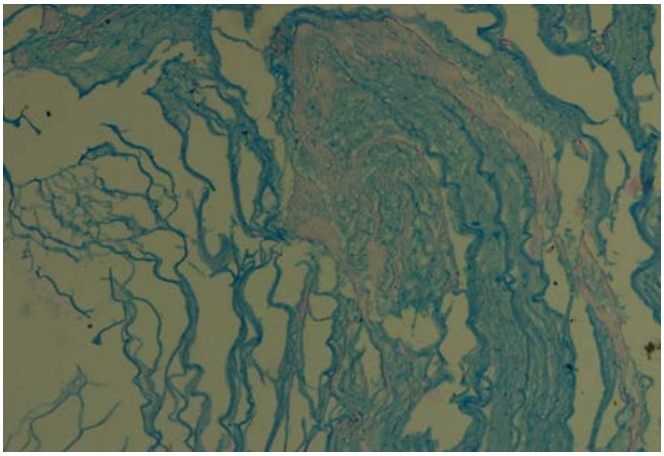

Figure 2. Histological sample stained with Alcian Blue, seen at intermediate magnification (×10) showing peritoneal mucinous neoplasia (Dr Ouattara S).

The patient was referred to the oncology department. Further assessment was carried out in February 2019. A thoraco-abdominopelvic CT scan did not find specific liver lesion, peritoneal effusion and thickened peritoneum. Biologically, there was hypochromic microcytic anemia with a hemoglobin level of 8.2 g/dl.

The patient received treatment based on Tenofovir tab 300 mg: 1 tab/day; of 2 bags of globular concentrates and two courses of chemotherapy based on cisplatin-doxorubicin-cyclophosphamide from 02 April 2019 to 02 May19.

The evolution was stationary with persistent ascites and anemia. For financial reasons, the patient was unable to continue the chemotherapy courses. In 2021, he presented with an altered general condition and oedemato-ascitic syndrome, and underwent a further course of chemotherapy. For financial reasons, he again had to interrupt his treatment and finally died in 2023.

3. Observation 2

A 51-year-old, female patient, housewife, was seen in a hepato-gastroenterology consultation in August 2016 for abdominal distension that had been evolving for three months without accompanying signs.

On physical examination, the patient was found to be in World Health Organization stage II general condition, with conjunctival and mucosal pallor, and ascites, the puncture of which yielded a gelatinous, hematic fluid. Chemical and bacteriological examination of the ascites fluid revealed the presence of a few leukocytes and red blood cells, but no pathogenic germs. Cytopathological examination of the ascites fluid revealed no cells suspicious of malignancy. Abdominal ultrasonography suggested peritoneal carcinosis, and abdominopelvic CT revealed partitioning membranes with varying degrees of enhancement after gadolinium injection, suggesting a tumoral process.

Biologically, there was microcytic hypochromic anemia with hemoglobin 9.1 g/dl, hyperleukocytosis (10,220/mm^3) with 74% neutrophils and thrombocytosis (782,000/mm^3). Alpha-feto-protein, transaminases, prothrombin level, protidogram, urea, creatininemia, blood ionogram and blood glucose returned normal.

The cytobacteriological study of the ascites fluid showed a viscous, purulent appearance with numerous red blood cells and leukocytes (59% neutrophils and 41% lymphocytes), with no germs found.

The September 2016 polymerase chain reaction indicated exploratory laparotomy. This allowed evacuation of a gelatinous substance with lumps occupying the peritoneal cavity, appendectomy and left oophorectomy. Pathological examination revealed pseudomyxoma peritonei secondary to a mucinous cystadenocarcinoma of the ovary and appendix.

She received six courses of adjuvant Paclitaxel-carboplatin chemotherapy.

The clinical and biological evolutions were favorable, but on the follow-up CT scan, moderate hepatomegaly and ascites of low abundance were noted. Given the persistence of ascites, tumour reduction surgery consisting of a total hysterectomy was performed in June 2017. Pathological examination revealed squamous metaplasia of the cervix and chronic endometritis. She subsequently underwent three courses of adjuvant chemotherapy. A thoraco-abdominopelvic CT scan performed in March 2018 returned normal.

4. Discussion

The cost of treatment was a limiting factor. Approximate prices were: 305 euros for surgery, 686 euros for six courses of chemotherapy, 77 euros for abdominal CT scan with injection, 30 euros for abdominopelvic ultrasound, 23 euros for anatomopathological study of a biopsy specimen; a total of 1121 euros. Patients had to pay for everything. This contributed to a delay in treatment, especially for patient No. 1, given that the average salary of a security guard in Burkina Faso is 77 euros.

This study shows that the diagnosis of pseudomyxoma is often delayed in our context. For the first patient, the diagnosis of certainty was made two years after the onset of symptoms, and for patient n˚2, five months after several consultations in different centers. For the first patient, the first ultrasound scan suspected a pseudomyxoma peritonei two years before the diagnosis of certainty was made. The existence of a specialized multidisciplinary structure for the management of rare diseases of the peritoneum might have enabled earlier diagnosis, thereby decreasing the cost of treatment.

Our patients were aged 60 and 51. According to the literature, age at diagnosis varies between 20 and 80 years [1]. They were one man and one woman.

The reason for consultation in our patients was abdominal distension. The symptomatology of pseudomyxoma peritonei, as emphasized by many authors, is not very specific, and it is often abdominal distension, the most common symp-

tom, that leads to the diagnosis of pseudomyxoma peritonei.

The abdominal distension, the pseudappendicular syndrom and the digestive disorders were the most frequent reasons for consultation [9]-[11].

On imaging, the first patient's initial abdominopelvic ultrasound suspected a pseudomyxoma peritonei. In both our patients, no initial abdominal CT scan suggested the diagnosis. Abdominal ultrasonography, according to Ben Salah in Morocco, had evoked the diagnosis in nine out of 15 cases [12]. The study of a greater number of cases in our context could enable us to adopt the proposal that, in the presence of gelatinous ascites with an ultrasound appearance suggestive of pseudomyxoma peritonei, laparoscopy could be proposed, thereby reducing costs and delays in management.

In the first patient, ascites initially had a lemon-yellow appearance, rich in proteins and leukocytes (700/mm^3 predominantly lymphocytes). This suggests tuberculinization of the ascites fluid. Even though the ascites persisted after anti-tuberculosis treatment, the cellularity had dropped to 16/mm^3. The patient's ascites was viscous and purulent, rich in red blood cells and leukocytes, with a slight predominance of neutrophils.

Laparotomy revealed gelatinous ascites in both patients. Both underwent appendectomy, with evacuation of the gelatinous masses, while the second patient underwent left oophorectomy and later total hysterectomy, with removal of the left adnexa.

Anatomopathological study concluded that patient n˚1 had a WHO grade I (2010) pseudomyxoma peritonei and patient n˚2 a pseudomyxoma peritonei secondary to an ovarian and/or appendicular mucinous cystadenocarcinoma.

Our patients received adjuvant chemotherapy: doxorubucine-cisplatin-cyclophosphamide for one and carboplatin-placlitaxel for the other. None received hyperthermic intraperitoneal chemotherapy. However, the evolution was initially stationary for the first patient, then he finally died, and favorable for the other. Patients suffering from pseudomyxoma peritonei should ideally undergo complete cytoreduction surgery and hyperthermic intraperitoneal chemotherapy in a referral center. The 5-year survival rate is 90%, and the 10-year rate 85% [10] [13]. We do not yet have a specialized center for the treatment of cancers, particularly pseudomyxoma. In the three-year follow-up, we had no recurrence in the second spatient, and the first patient died. It would be advisable to set up a committee for reflection and follow-up, which could propose the best possible and feasible management in our context.

5. Conclusion

Pseudomyxoma peritonei remains a very rare anatomo-clinical entity in our context, with often delayed diagnosis and therapeutic difficulties. Its therapeutic approach today recommends cytoreduction surgery and intraperitoneal chemotherapy in a specialized center. Management in our context is hampered by financial difficulties and the absence of a suitable management framework. These difficul-

ties could be alleviated to some extent by establishing a framework for reflection and care of this condition adapted to our country's resources.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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