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Case Report

A Brief Overview of Our Case Report and other Previous Studies About Pseudomyxoma Peritonei in Pregnant Case

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Abstract:

Pseudomyxoma peritonei (PMP) is a rare clinical condition characterized by mucinous ascites and peritoneal implants, typically originating from the appendix or ovary. Its occurrence during pregnancy is exceedingly rare and presents significant diagnostic and therapeutic challenges. We present the case of a 36-year-old primigravida at 19 weeks gestation, presenting with progressive abdominal distension and jaundice. Imaging revealed a large pelviabdominal mass, initially suspected to be a degenerating fibroid. Exploratory laparotomy revealed a massive mucinous tumor. Histopathology confirmed pseudomyxoma peritonei. The patient underwent surgical management with a successful outcome. This case highlights the diagnostic dilemmas and surgical considerations in managing PMP during pregnancy.

In conclusion, pseudomyxoma peritonei in pregnancy is a diagnostic and management challenge. High clinical suspicion, careful imaging, and timely surgical intervention are crucial. This case adds to the limited body of literature and highlights the importance of considering rare diagnoses in atypical presentations.

Keywords: large pelviabdominal mass; pseudomyxoma peritonei; pregnancy; dpam, pmca

Introduction

Pseudomyxoma peritonei (PMP) often called ‘‘jelly belly’, is a rare cancer that generally presents as multifocal mucinous tumors in the abdominal cavity causing increased abdominal girth, pain and pressure on internal organs due to the accumulation of large amounts of mucinous tumor (Patrick-Brown et al., 2020; Garg et al., 2024). Pseudomyxoma peritonei (PMP) is a form of peritoneal malignancy. It originates from a perforated appendiceal epithelial tumor and affects 22 individuals per million worldwide (Patrick-Brown et al., 2020). The cancer spreads along the peritoneum, a thin layer that protects the abdominal organs, and can involve the surface of all abdominal organs. Without treatment, PMP is a fatal condition (Ionescu et al., 2024). The unlimited multiplication of peritoneum cells can fill the space needed in the abdomen for normal gastrointestinal functioning, leading to compression of bowel organs, disruption of their functioning, and starvation (Taher et al., 2024).

Initial presentation varies and consists of unspecified signs and symptoms that relate to the progression of the disease (García et al., 2019). These include increased abdominal girth, an appendicitis-like syndrome, a new-onset hernia, presence of a pelvic mass, or non-specific abdominal or pelvic pain (Awad et al., 2024). Progressive accumulation of mucinous material gradually fills and can compress vital organs within the peritoneal cavity, which can result in abdominal distention, ascites, bowel obstruction, and nutritional compromise (Arslan, 2023).

The prognosis with the histological features. These included disseminated peritoneal adenomucinosis (DPAM) and peritoneal mucinous carcinomatosis (PMCA), (Viloria and Amosco, 2023). DPAM consisted of peritoneal lesions composed of abundant extracellular mucin containing scant simple to focally proliferative mucinous epithelium with little cytologic atypia or mitotic activity, with or without an associated appendiceal mucinous adenoma (Ye and Zheng, 2022; Arrington, 2022). PMCA was composed of peritoneal lesions containing more abundant mucinous epithelium with the architectural and cytologic features of carcinoma, with or without an associated primary mucinous adenocarcinoma (Martín-Román et al., 2021).

The most common treatment for PMP is cytoreductive surgery (CRS) followed by hyperthermic intraperitoneal chemotherapy (HIPEC) (Papantoni et al., 2021). CRS entails removing the peritoneum and other affected tissues. CRS is an extensive and complex surgery. HIPEC is a therapy that uses chemotherapy that is applied directly to the abdomen (Karimi et al., 2024). Patients with PMP experience various stressful and traumatic events, including diagnosis with a rare disease, treatment with extensive and

complex surgery, admission to intensive care for an average of 5 days, and then on a surgical ward for an average of 3 weeks (Taher et al., 2024).

For patients that cannot be cured by surgery, no effective treatments exist, and since PMP is a slow-growing cancer, patients may live for many years with active disease and worsening symptoms, presenting a substantial burden on the health-care system (Patrick-Brown et al., 2020). Estimation of prevalence is therefore essential for resource allocation. Prevalence estimates depend on the incidence rate, which for PMP have been widely quoted as approximately 1–2 people per million (Li et al., 2024).

Case Presentation

A 36-year-old primigravida at 19 weeks gestation presented with acute abdominal distension, progressive discomfort, and jaundice. She had no previous surgical or medical history but was a known case of uterine fibroids. Physical examination revealed a markedly distended abdomen with a palpable pelviabdominal mass. Ultrasound and Magnetic Resonance Imaging (MRI) showed a large heterogeneous lesion measuring approximately 22×16×11 cm, suspected to be a degenerating fibroid, see figure 1a, b. Given the worsening symptoms, an exploratory laparotomy was performed, revealing a massive mucinous mass. The uterus was gravid and displaced by the lesion. Resection of the tumor was achieved, and samples were sent for histopathology, which confirmed pseudomyxoma peritonei.

Post operative course pregnancy follows up and delivery with outcome details

In our case there were no significant events observed in the postoperative course or pregnancy follow-up. Also, the patient was doing well and had no complaints; at the same time, the patient had received adequate analgesia, and she had passed urine and stool. Additionally, our case had tolerated an oral diet and mobilized adequately without assistance. Regarding the baby, it was admitted to the neonatal intensive care unit (NICU).

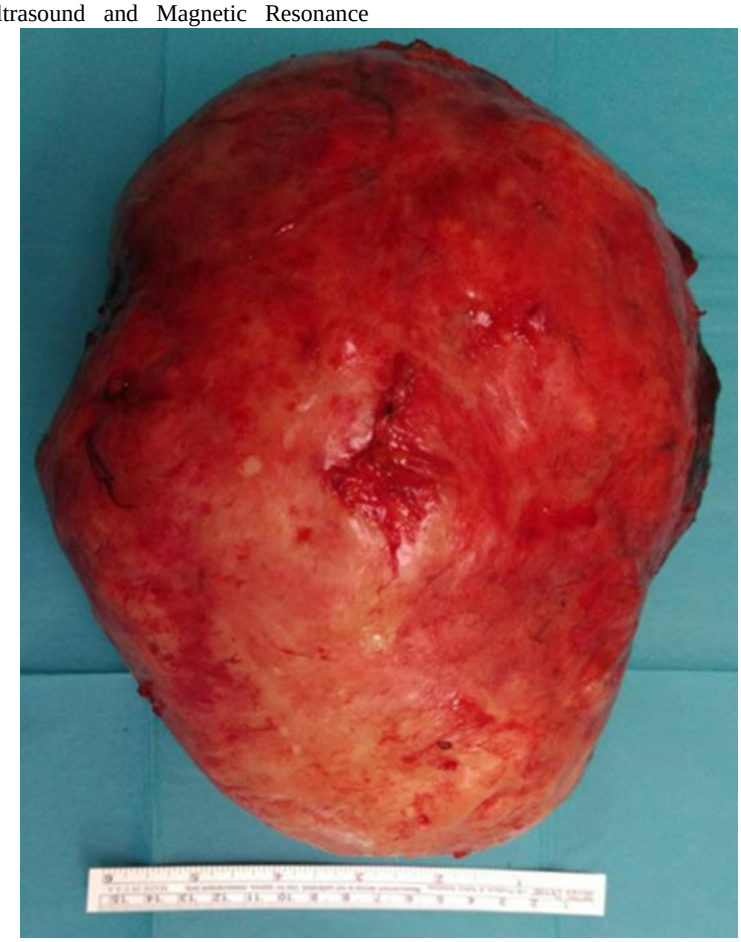

Figure 1a: Gross specimen of the resected mass showing a large, encapsulated lesion with a smooth, glistening surface and focal areas of vascularity and hemorrhage. The mass measures approximately 25 cm in greatest diameter.

Discussion

Pseudomyxoma peritonei during pregnancy is extremely rare, and diagnosis can be delayed due to overlapping symptoms with pregnancy-related changes (Haase et al. 2009, Koyama et al. 2011). Imaging studies are limited due to fetal safety concerns, and definitive diagnosis often occurs intraoperatively (Sugarbaker, 2007).

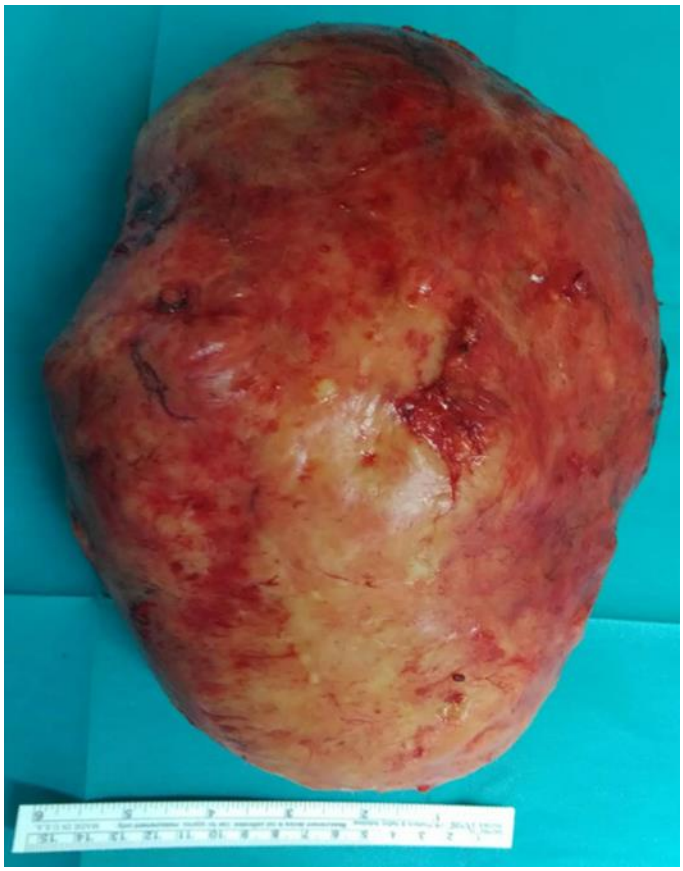

Figure 1b: Another view of the same specimen is taken under different lighting to highlight its lobulated external surface. The scale below the mass shows the large size and irregular contour.

In our case, the mass was initially misinterpreted as a fibroid. Surgical exploration and resection during the second trimester proved safe and effective (Esquivel and Sugarbaker, 2000). Histological evaluation remains the gold standard for diagnosis (Ronnett et al., 1995). multidisciplinary planning, including gynecologists, surgeons, and pathologists, is essential for optimal outcomes (Bevan et al., 2010). In another case report in Egypt, by Mousa et al., (2021), A 27-year-old primigravida, at 30 weeks of gestation, was transferred to our tertiary care obstetric center. Her Gynecologists performed an US guided ovarian cyst aspiration five days ago with no improvement. Three-dimensional (3D) US showed a huge multilocular left ovarian cyst (20 cm) with smooth outline and contents showing heterogeneous echogenicity. No solid components or papillary projections were detected in the cyst. It also showed pelvi-abdominal ascites reaching the level of hepatorenal pouch. The patient had smooth postoperative recovery and was discharged on day 5 postoperative after her lab results approached normal values. She was counselled about comprehensive surgical staging with fertility preservation versus complete staging laparotomy (Maltaris et al., 2006, Marpeau et al. 2008).

Specimens were sent for tissue histopathology and fluid cytology examination. No intra-abdominal organs or peritoneal masses that might require cytoreduction were detected intraoperatively (Desai et al., 2014). However, in our case, Gross specimen of the resected mass showing a large, encapsulated lesion with a smooth, glistening surface and focal areas of vascularity and hemorrhage. Ultrasound and Magnetic Resonance Imaging (MRI) showed a large heterogeneous lesion measuring approximately 22×16×11 cm, suspected to be a degenerating fibroid. Indeed, 1% to 2% of pregnant women have an adnexal mass discovered on ultrasonography [9]. A review of the literature found only seven other cases of pseudo myxoma peritonei diagnosed during pregnancy (Table 1).

Case	**Our case**	**Ben Abdu2009**	**Erika Haase 2009**	**Shinsuke Koyama 2010**	**Z. Manan 2010**	**Sofia Jayi 2012**	**Mousa et al. 2021**	**Basso et al. 2022**
Age/yr	36 years	36 years old	30 years old	34 years old	41 years old	35 years old	27-years-old	34 years old
Diagnosis	Sepsis and disseminated Intravascular coagulopathy	Well-differentiated appendix mucinous adenocarcinoma with pseudomyxoma peritonei	Well-differentiated appendix mucinous adenocarcinoma with pseudomyxoma peritonei	Transverse colon mucinous adeno Carcinoma with ovarian metastasis and pseudomyxoma peritonei	Well-differentiated appendix mucinous adenocarcinoma with pseudomyxoma peritonei	Mature teratoma and mucinous borderline ovarian tumor with pseudomyxoma peritonei	Sepsis and disseminated Intravascular coagulopathy	Well-differentiated appendix mucinous adenocarcinoma, with pseudomyxoma peritonei

Clinical presentation	Acute abdominal distension, progressive discomfort, jaundice	During cesarean section	Accidental finding in routine pre Natal ultrasound	Accidental finding in routine pre Natal ultrasound	During cesarean section	Abdominal pain	Acute Abdominal distension, discomfort and constipation	Abdominal pain
GA at presentation	19 weeks	At delivery	17 weeks	24 weeks	At delivery	22 weeks	30 weeks	29 weeks
Management during pregnancy	Surgical exploration and resection during the second trimester proved safe and effective		Laparotomy with right salpingooophorectomy, appendectomy, Omental biopsy	Pararectus incision, Appendicectomy		Salpingooophorectomy, hysterectomy, omentectomy, appendicectomy, iliac lymphadenectomy	Huge multilocular left ovarian cyst (20 cm)	Laparotomy with right adnexa Tomy, omentectomy, Appendicectomy
Management delivery	Cesarean section at 36 weeks	Cesarean section for failure to induce labor 40 GW	Labor induction Vaginal delivery at 35 weeks	Cesarean section at 34 weeks	Labor induction for Macrosomia Cesarean section for failure to induce labor 40 GW	Pregnancy interruption at 22 WG	classical caesarean section (CS)	Labor induction Vaginal delivery at 37 weeks
Treatment	Complete cytoreductive surgery with HIPEC	Appendicectomy, right colectomy with HYPEC during cesarean	Complete cytoreductive surgery with HIPEC and systemic chemotherapy (8 cycles with Xeloda and oxaliplatin)	Complete cytoreductive surgery with bilateral salpingooophorectomy, partial resection of the transverse colons follow by chemotherapy (FOLFOX and bevacizumab)	Appendicectomy, mucus resection	Trial of conservative management with parenteral antibiotics, intravenous (IV) fluids, hemodynamic and respiratory support		Complete cytoreductive surgery with HIPEC
Follow-up	Recurrence, 1 month after surgery		5 years no recurrence	6 months progressing	Lost of follow up		Recurrence, 4 weeks postoperative	Recurrence, 14 months after surgery

Table 1: Cases of pseudomyxoma peritonei during pregnancy.

For this review, we included all studies on pseudomyxoma peritonei in pregnant women. We performed the search among articles in the PubMed database. The mesh terms used were "pseudomyxoma peritonei" and "pregnant women" or "pregnancy." The search was extended to include the last 20 years. All types of studies were included. Titles and abstracts were read, and if the articles were relevant to the subject, they were selected. Pseudomyxoma peritonei syndrome is a peritoneal gelatinous disease associated in most cases with appendiceal adenocarcinoma intraperitoneal rupture. The discovery of an ovarian mass in pregnancy is often incidental during an obstetrical ultrasound (US) scan, as in the cases of E. Haase (2009) and S. Koyama (2011).

Therefore, it is important that the ovaries be examined during the first trimester with US scan. AUS scan is the first-line exam for the characterization of an ovarian mass. Pelvic MRI is useful during pregnancy in cases of uncertain diagnostics on US scans. It is the exam of choice when the pregnancy is advanced or when the tumor is large, as in our case [10]. The French recommendations for the management of ovarian masses and malignant tumors diagnosed during pregnancy allow us to propose management according to the diagnosis suspected on imaging and the term of the pregnancy, this is summarized in (Figs. 2) [10, 11, 15].

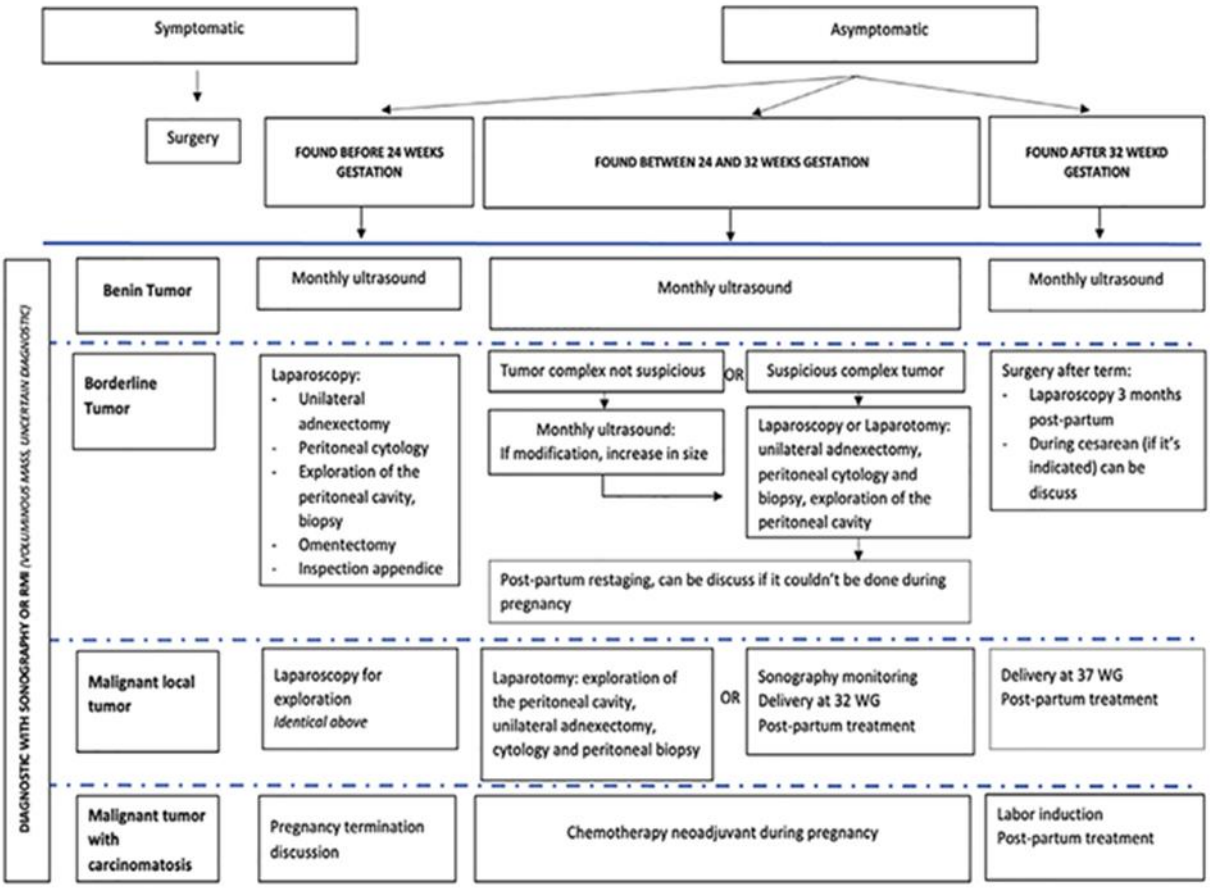

Figure 2: Suggested management strategy for ovarian mass discovery during pregnancy.

Of course, this management must be adapted according to the patient's age, the anatomopathological final diagnosis, the desire of future pregnancy and the current pregnancy. In the literature review, three cases of PMP were diagnosed before 28 weeks gestation. The other two cases were diagnosed at the time of Caesarean section [5, 6]. In E. Haase [4] and S. Koyama [8] cases, the pregnancy was continued until 34/35 weeks gestation where the risk to the premature infant is quite low. In the case of Sofia Jayi 2012 [7], the pregnancy was terminated at 20 weeks gestation to perform cytoreduction. It was patient wishes.

The prognosis of PMP depends on two main factors: the disease's grade and the cytoreduction's quality. The PMP can be classified as low grade or high grade PMP according to the 2010 WHO classification [1]. Low-grade appendicular PMP has a slower and less aggressive course than high-grade PMP [12, 13], with a better prognosis and a lower risk of recurrence. The low grade PMP natural history is very slow and progressive. For this reason, it seemed appropriate to induce delivery at 37 weeks gestation and wait 1 month postpartum before performing cytoreduction and HIPEC.

According to her, for high-grade PMP diagnoses at first or second gestation trimester the pregnancy termination must be considered. In the low-grade PMP because of the very slow progression disease, we can wait until 35 weeks whatever the diagnostic term. The delay in management induced by continued pregnancy certainly did not have a significant impact on the development of PMP. The PCI score in antepartum and postpartum remained quite the same (PCI at 21 in antepartum, without pelvis exploration, compared to 27 in postpartum after pouch of Douglas" +3, and left parametria peritoneum +2 exploration). Cytoreduction was macroscopically complete, which is the main prognostic factor. Finally, the 5-year survival of patients treated with cytoreduction and CHIP for low-grade PMP is 63%, compared with 23% for high grade PMP [12].

Conclusion

Pseudomyxoma peritonei in pregnancy is a diagnostic and management challenge. High clinical suspicion, careful imaging, and timely surgical intervention are crucial. This case adds to the limited body of literature and highlights the importance of considering rare diagnoses in atypical presentations.

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