CASE REPORT

Peritoneal inclusion cysts in an adolescent female: A case report and review of the literature

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Abstract

Several studies have shown that most peritoneal inclusion cysts (PIC) cases occur almost exclusively in women of childbearing age and patients who have had abdominal or pelvic surgery. We report a case of PIC diagnosed in a 19 years old single adolescent female with chronic pelvic pain and secondary amenorrhea with no prior history of abdominal surgery. A multilocular abnormality in the pelvis and a septated pelvic fluid encompassing the ovaries and extending to the right iliac fossa was revealed by ultrasound images and Magnetic Resonance Imaging (MRI), respectively. She was treated with broad-spectrum antibiotics, and after symptoms persisted, a laparoscopic exploration that resolved all symptoms was performed. After histological analysis, she was diagnosed with peritoneal inclusion cysts**.** PICs are a rare non-ovarian cause of cystic pelvic lesions and should be considered in its differential diagnosis. Furthermore, diagnostic laparoscopy can be performed when no precise radiological diagnosis is obtained. Surgical excision of all visible cyst walls remains the best-recommended treatment. *(Afr J Reprod Health 2023; 27 [7]: 127-132).*

Keywords: Peritoneal inclusion cysts, chronic pelvic pain, diagnostic laparoscopy, adolescent female, diagnostic challenge

Résumé

Plusieurs études ont montré que la plupart des cas de kystes d'inclusion péritonéaux (PIC) surviennent presque exclusivement chez les femmes en âge de procréer et les patients ayant subi une chirurgie abdominale ou pelvienne. Nous rapportons un cas de PIC diagnostiqué chez une adolescente célibataire de 19 ans souffrant de douleurs pelviennes chroniques et d'aménorrhée secondaire sans antécédent de chirurgie abdominale. Une anomalie multiloculaire du bassin et un liquide pelvien cloisonné englobant les ovaires et s'étendant jusqu'à la fosse iliaque droite ont été révélés respectivement par des images échographiques et par imagerie par résonance magnétique (IRM). Elle a été traitée avec des antibiotiques à large spectre et, après la persistance des symptômes, une exploration laparoscopique qui a résolu tous les symptômes a été réalisée. Après analyse histologique, elle a été diagnostiquée avec des kystes péritonéaux d'inclusion. Les PIC sont une cause non ovarienne rare de lésions pelviennes kystiques et doivent être prises en compte dans son diagnostic différentiel. De plus, la laparoscopie diagnostique peut être réalisée lorsqu'aucun diagnostic radiologique précis n'est obtenu. L'exérèse chirurgicale de toutes les parois visibles du kyste reste le traitement le plus recommandé. *(Afr J Reprod Health 2023; 27 [7]: 127-132).*

Mots-clés: Kystes d'inclusion péritonéaux, douleur pelvienne chronique, laparoscopie diagnostique, adolescente, défi diagnostique

Introduction

Peritoneal inclusion cysts (PICs) are fluid-filled benign multilocular cysts found between intraperitoneal adhesions[1]. Most often, it affects females with healthy ovaries[2]. The peritoneum's ability to absorb fluid diminishes when its integrity has been compromised, as might happen after surgery, trauma, or inflammation brought on by conditions like pelvic/abdominal inflammatory diseases or endometriosis, resulting in complex cystic mass due to a buildup of trapped ovarian fluid[3]. Along with endometriosis, inflammatory bowel disease, and pelvic inflammatory disease, PIC can also be observed in those conditions[4-6]. Therefore, peritoneal inclusion cyst formation that is consistent with the presentation of this lesion after puberty requires both functionally active ovaries and adhesions[7]. Several studies have shown that most PIC cases occur almost exclusively in women of

childbearing age and patients who have had abdominal or pelvic surgery[1,4,8-10]. Very few cases of PIC in adolescents have been previously described[7,8]. All cases, however, had histories of previous abdominal surgery.

Although some studies indicated PICs are treated with dienogest, which has shown to be quite successful for this purpose[11] , the matter of PIC treatment and diagnosis still needs additional significant studies[12]. We here present a case of PIC diagnosed in an adolescent female with no prior history of abdominal surgery.

Case Report

A 19 years old single female was referred to our outpatient clinic at King Faisal specialist hospital and research center in Al- Madina,with chronic pelvic pain and secondary amenorrhea. She had no history of gynaecological disease or abdominal surgery. A multilocular abnormality in the pelvis, measuring 10.57×5.48 centimetres in diameter, was revealed by ultrasound images (Figure 1). Bilateral polycystic ovaries were shown, with cystic regions that were both homogeneous and partially dense. A second MRI scan of the pelvis was conducted to learn more about the mass's precise location. The results revealed a septated pelvic fluid encompassing the ovaries, measuring 11 x 9 x 7.7 cm, extending to the right iliac fossa (Figure 2).

A pelvic inflammatory disease was suspected, and she was placed on broad-spectrum antibiotics for 14 days. However, symptoms persisted despite the treatment. MRI was repeated after one month to reassess the case, and findings showed a significant amount of free fluid in the pelvis, raising the suspicion of a peritoneal inclusion cyst. An ultrasound-guided cyst aspiration was performed, which failed due to severe pain, and marked resistance at the wall of the cystic lesion resulting in a lack of fluids during aspiration.

Further laboratory investigations were ordered, including brucella serology, Tuberculoais (TB) Quantiferon test, Echinococcosis granulosis serology, and serology for hydatid disease. The pelvic aspirate was also sent for Gram staining, culture and sensitivity, Acid-Fast Bacillus smear (AFB), Mycobacterium TB PCR and Mycobacterium TB culture and all results came out negative. The patient was then considered for exploratory laparotomy and cyst excision. The procedure revealed a multicystic lesion in the cul-de-sac, stretching from the inferior to the right Waldeyer's fossa, independent of the ovaries, the sigmoid colon, and the rectum (Figure 3).

There were no intraoperative or postoperative complications, and all cysts were successfully removed. Histological and cytological examination of the multiple cystic biopsies revealed no signs of cancer, confirming the PIC diagnosis. There were no further postoperative problems, and the patient's abdominal symptoms had resolved by the 6-week follow-up. Further follow-up is scheduled at 6 and 12 months.

Discussion

PICs which develop from the peritoneal mesothelium, are a rare non-ovarian source of pelvic cysts[13]. Though thought to only be neoplastic, research suggests they are reactive proliferation in response to a peritoneal shock caused by endometriosis, pelvic inflammatory illness, or abdominal surgery[4,8,10,13]. Contrarily, it is interesting to note that the patient, in this case, did not present with these common risk factors resulting in a diagnostic challenge. Most PICs are discovered by chance, but when they produce symptoms, they often manifest abdominal distension and pain[14]. Low back or pelvic pain, as presented in this case, is according to studies of the typical presenting symptom of PIC[7,15].

PIC can present a wide range of symptoms, most times leading to inaccurate diagnosis. It is also hard to identify them in imaging studies because of the difficulty distinguishing them from an ovarian tumour. In this case, imaging studies revealed a multilocular abnormality in the pelvis, measuring 10.57×5.48 centimetres in diameter (Figure 1), suggesting a Multi loculated cystic lesion. PICs manifest radiographically as multilocular cystic lesions containing fluid and occasionally hemorrhagic components, often taking on a spider web or honeycomb appearance[16,17].

Together with ultrasound imaging, magnetic resonance imaging (MRI) is utilised to help further characterise complex cystic masses and reveal anatomic relationships with the surrounding organs[10]. Usually, effective treatments involve interventional radiology or surgical resection (IR),

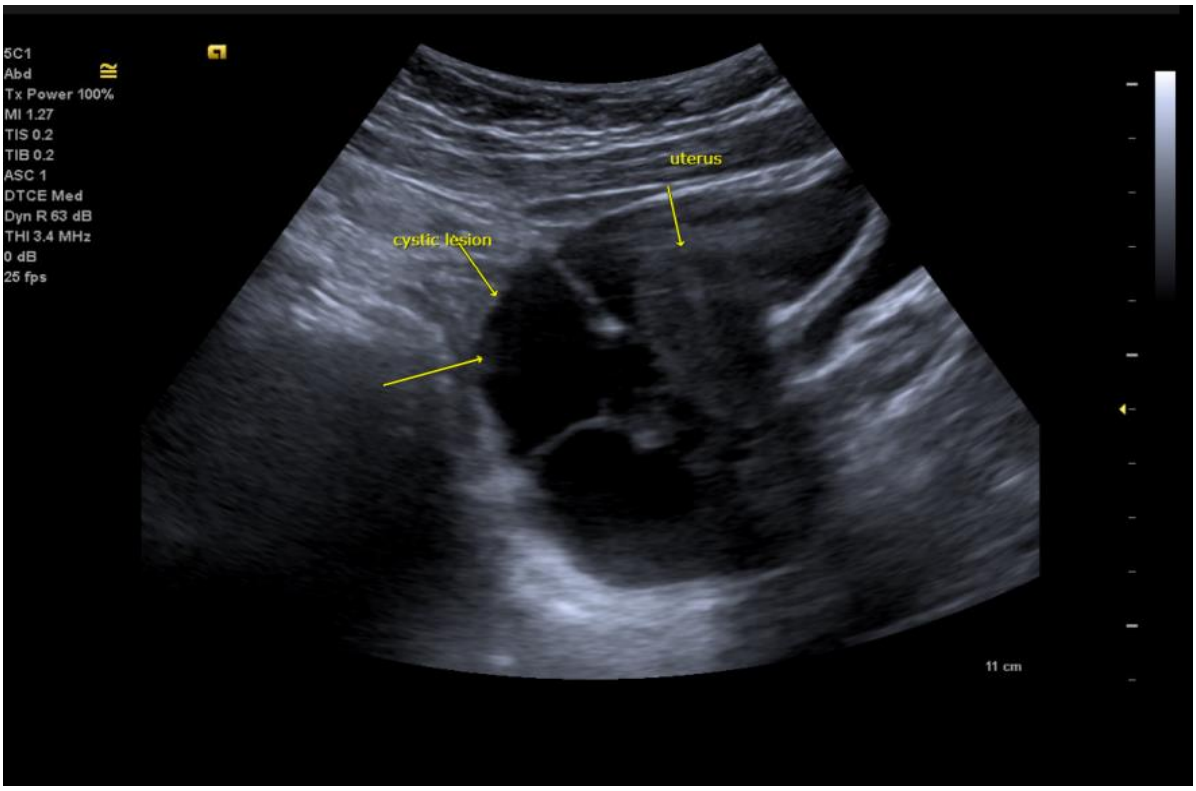

Figure 1: Ultrasonography showing a multinodular abnormality of 10.57×5.48 cm in diameter

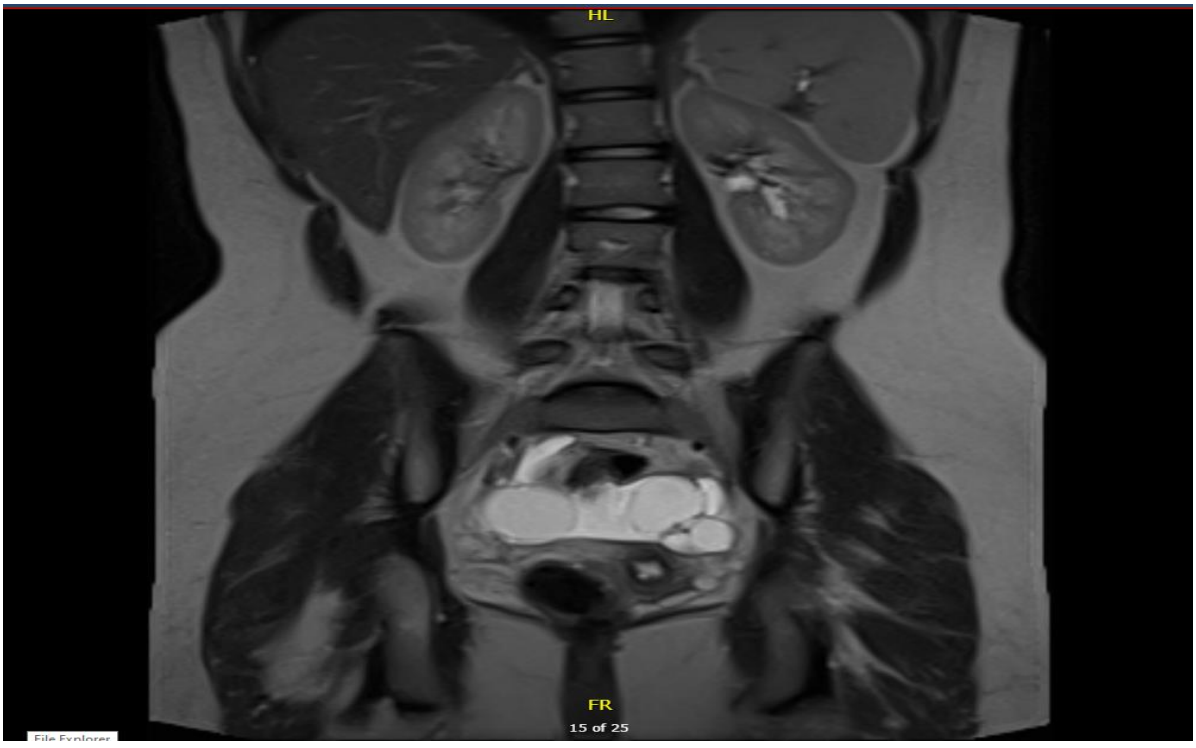

Figure 2: MRI showing a Septated pelvic fluid encompassing the ovaries, measuring 11 x 9 x 7.7 cm, extending to the right iliac fossa

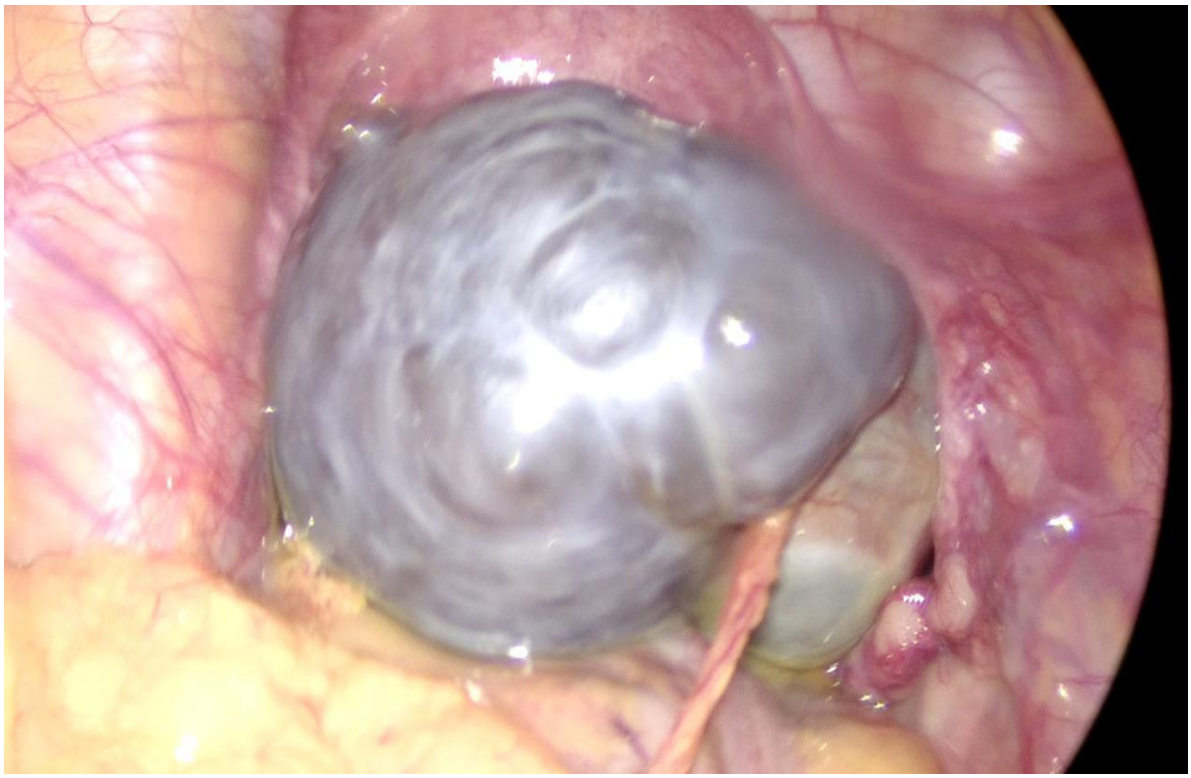

Figure 3A: Exploratory laparotomy showing the largest inclusion cyst

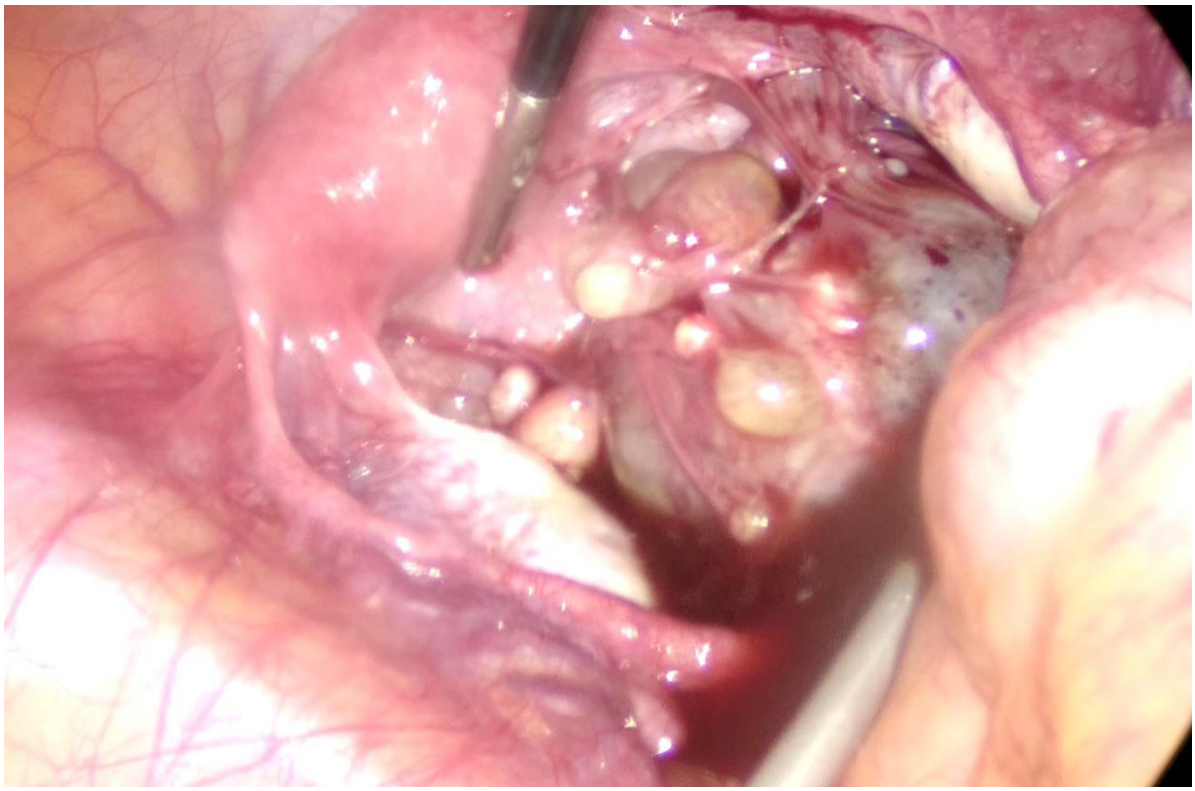

Figure 3B: Exploratory laparotomy showing several small cysts found on the base behind the largest cyst

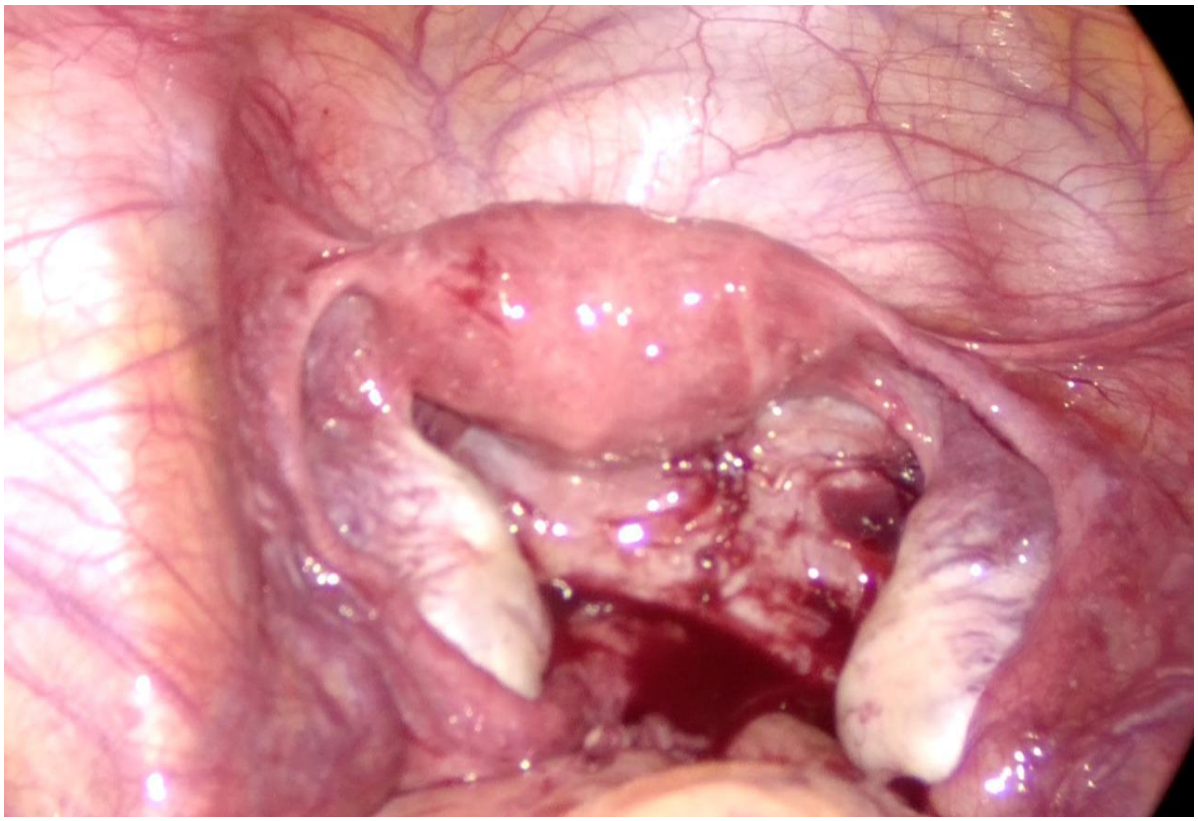

Figure 3C: Exploratory laparotomy showing the pelvis after the removal of all the inclusion cysts

treating PIC can be made with the help of ultrasound imaging alone[18]. However, as seen in this case, the size and intricacy of the lesions, as well as the uncertainty regarding their source, necessitated additional imaging. Although the MRI result was of little help, a further Ultrasound-guided aspiration also yielded no beneficial result.

Definitive diagnoses were acquired through the results from a histological examination after diagnostic laparoscopy showing the procedure can be performed when radiological diagnosis yields an unclear result. Due to its challenging diagnosis, PIC often results in delayed treatment [10,16]. In this case, a 14 days treatment with broad-spectrum antibiotics yielded no result, but all symptoms were resolved after the surgical excision of the cyst. Therefore the surgical excision of all visible cyst walls is the most effective treatment of PIC, consistent with several studies[4,7-10].

Although other treatment options such as imaging observation and follow-up in asymptomatic patients[19], sclerotherapy, Image-guided transvaginal or percutaneous fluid aspiration, pain medications, and GnRH analogues[20], levonorgestrel-releasing intrauterine system[1], hormonal therapy to suppress ovulation[1,10], have all been recorded. However, the success rates of these non-surgical options are far lower than those of surgery[4].

In conclusion, this case presents an adolescent female diagnosed with PIC, and findings reveal that PICs are a rare non-ovarian cause of cystic pelvic lesions and should be considered in its differential diagnosis. Furthermore, diagnostic laparoscopy can safely be performed when there is no precise radiological diagnosis. Surgical excision of all visible cyst walls remains the best-recommended treatment, although other management options can be considered in an asymptomatic patient.

Data availability

Data used to write this case report is accessible through contact with the corresponding author, Amal Yaseen Zaman, email: amalyzaman@hotmail.com

Consent

Written informed consent was taken from the patient for participation in the study and publishing the results.

Competing interests

The authors declare that they have no competing interests.

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