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Case Report & Literature Review

Hematocolpos secondary to imperforate hymen presenting with acute abdominal pain and misdiagnosed as appendicitis: a case report and literature review

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ABSTRACT

Imperforate hymen is a rare congenital malformation that usually presents in adolescence with primary amenorrhoea and pain or pressure-related symptoms from haematocolpos. We present the case of an 11-year-old girl who presented as a surgical emergency with acute abdominal pain. A laparoscopy was performed for suspected appendicitis, but she was subsequently found to have a haematocolpos secondary to imperforate hymen. This case illustrates the challenges in making the diagnosis. The literature contains numerous cases where the diagnosis has been delayed, and we discuss those that also presented as possible appendicitis. These cases highlight the importance of awareness of the condition amongst general surgeons and emergency physicians, and the role of external genital examination and ultrasonography prior to surgical intervention.

Introduction

Imperforate hymen is a rare condition, resulting when the hymen, a thin membrane at the junction of the sinovaginal bulb and urogenital sinus, fails to perforate during fetal development. At menarche, blood collects behind the imperforate hymen, forming hematocolpos, and leading to the typical presentation of cyclical pelvic pain and primary amenorrhoea. We describe a case that presented more acutely, mimicking a surgical emergency, and resulted in an unnecessary laparoscopy and delayed diagnosis. A literature review of similar cases is presented along with important clinical lessons.

Case Presentation

An 11-year-old girl presented as an emergency with a two-day history of increasingly severe, central abdominal pain. There were no associated urinary or bowel symptoms. There was no background of previous abdominal or pelvic pain. Her abdomen was soft, with tenderness centrally and in the right iliac fossa. There was no guarding or rebound tenderness. Inspection of the external genitalia was not considered. Pubertal development was not formally assessed. Her leucocyte count was normal but C-reactive protein was elevated at 59 mg/L. Urine microscopy and urinalysis were negative. An ultrasound scan was not requested. Acute appendicitis was considered as a possible diagnosis and she was admitted to the surgical department.

Her pain did not settle with 24 hours of conservative management and she underwent a diagnostic laparoscopy for presumed appendicitis. The findings at laparoscopy consisted of a normal-looking appendix, normal ovaries, bilaterally distended fallopian tubes, a distended uterus and approximately 100ml of old blood in the peritoneal cavity (Figures 1, 2, 3). Laparoscopic appendicectomy, with evacuation of hemoperitoneum and saline lavage, was performed. External genital assessment was still not undertaken.

The gynaecologist on-call was requested for an opinion on the presumed laparoscopic finding of retrograde menstruation. Post-operatively, a gynaecological history was taken. The patient gave no history of menarche and she was not sexually active. She was noted to have Tanner

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stage 1 breast and pubic hair development. A trans-abdominal pelvic ultrasound scan showed hematocolpos and hematometra.

A gynaecological examination under anaesthetic was performed and demonstrated a bulging, blue-tinged imperforate hymen. A cruciate incision was made in the hymen and 500ml of altered blood was drained from the vagina.

The patient made an uneventful recovery, with resolution of her pain. She was reviewed three months later by a gynaecologist with expertise in adolescent problems, and she reported a normal menstrual cycle with no further abdominal pain.

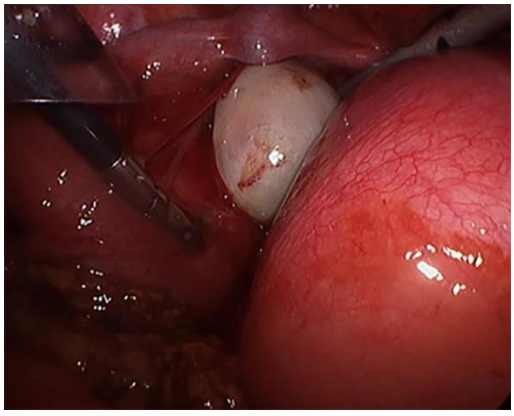

Figure 1: Laparoscopic photo showing distended uterus and dilated left fallopian tube with normal left ovary

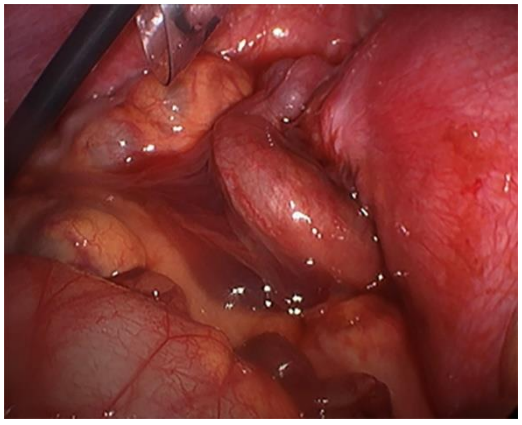

Figure 2: Laparoscopic photo showing dilated left fallopian tube with small amount of free blood in peritoneal cavity

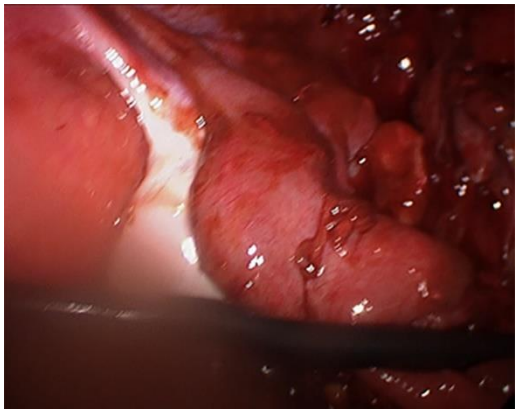

Figure 3: Laparoscopic photo showing dilated right fallopian tube with normal right ovary

Discussion

As this case demonstrates, doctors reviewing girls around the age of menarche with abdominal pain need to be aware that hematocolpos can present as a surgical abdomen. This includes emergency physicians, surgeons and gynaecologists. It is certainly not the first example of a patient with hematocolpos undergoing an inappropriate laparoscopy. In one case series, three out of 18 adolescents presenting with pain due to hematocolpos received unnecessary appendicectomies [1]. There are two further reported cases of adolescent girls with hematocolpos undergoing surgical procedures for suspected appendicitis: one a diagnostic laparoscopy and the other a McBurney laparotomy [2, 3]. In a further series of 13 cases, two had preliminary diagnoses of appendicitis, but ultrasonography diagnosed imperforate hymen prior to any operative procedure [4]. There is an additional case where appendicitis was strongly suspected in a patient who had refused genital examination [5]. The patient was booked for a laparoscopic appendicectomy, but imperforate hymen was diagnosed with ultrasonography en-route to the operating theatre. These cases all highlight the importance of gynaecological history taking, external genital examination and pelvic ultrasonography, in particular prior to any surgical intervention.

As breast development and the onset of adrenarche precede menarche, there is typically evidence of otherwise normal pubertal development at presentation, and in one case series of 13, this was true in all cases [6]. However, our case shows that one must consider imperforate hymen, even in the absence of clinical evidence of puberty. We were unable to find other examples of this in the literature.

There are numerous reports of delayed or missed diagnoses of imperforate hymen. One audit found that 6 out of 8 patients presenting via the emergency department, had the diagnosis missed on their first attendance [7]. Part of the problem may be a reluctance to perform external genital examinations, possibly due to embarrassment of the patient and physician [8]. In a series of 13 patients, genital examination was missing in two thirds [4].

Transabdominal emergency point-of-care ultrasound can be useful in facilitating a diagnosis in the emergency department [9, 10]. Another series of 13 cases of imperforate hymen, found that those diagnosed in the emergency department were significantly more likely to be admitted under gynaecology, receive a timely corrective procedure and be discharged sooner [10]. A timely diagnosis not only helps to alleviate symptomatology but is also important to reduce the risk of significant complications, such as acute urinary tract obstruction, tubal rupture, endometriosis, pelvic adhesions, and severe pelvic infection [6, 11-13].

In conclusion, an awareness of the varied presentations of imperforate hymen, including where it mimics a surgical abdominal emergency, is required for prompt diagnosis of the condition. Gynaecological history taking and external genital examination should be considered in girls presenting with abdominal pain and ultrasonography should be considered prior to surgery. This case also suggests that hematocolpos secondary to imperforate hymen can even occur in girls without other signs of puberty.

References

1. Nazir Z, Rizvi RM, Qureshi RN, Khan ZS, Khan Z (2007) Congenital vaginal obstructions: varied presentation and outcome. *Pediatr Surg Int* 22: 749-753. [Crossref]
2. Afshan N, Nargund A (2016) Unusual presentation of cryptomenorrhoea. *BJOG: An Int J Obstet Gynaecol* Conference: RCOG National Trainees Conference, NTC 2016. United Kingdom. 123: 31-32.

3. Nohuz E, Moreno W, Varga J, Tamburro S, Yanez M, et al. (2010) Imperforate hymen: one diagnosis can hide another. *Arch Pediatr* 17: 394-397. [Crossref]
4. Posner JC, Spandorfer PR (2005) Early detection of imperforate hymen prevents morbidity from delays in diagnosis. *Paediatr* 115: 1008-1012. [Crossref]
5. Patel RV, Campbell A, Njere I, Rejoo D (2013) Adolescent haematopyocolpos simulating appendicular abscess. *BMJ Case Rep*. [Crossref]
6. Ben Temime R, Najar I, Chachia A, Attia L, Makhlouf T, et al. (2010) Imperforate hymen: a series of 13 cases. *Tunis Med* 88: 168-171. [Crossref]
7. Logarajah V, Teng SS, Tyebally A (2012) What lies beneath! An audit of cases of imperforate hymen. *Proceedings of Singapore Healthcare*. Conference: SingHealth Duke-NUS Scientific Congress 2012. Singapore.
8. Lubsen-Brandsma MA, Salvatore CM (2008) Imperforate hymen; importance of inspection of the external genitalia [Article in Dutch]. *Ned Tijdschr Geneeskd* 152: 533-537. [Crossref]
9. Fischer JW, Kwan CW (2014) Emergency point-of-care ultrasound diagnosis of hematocolpometra and imperforate hymen in the pediatric emergency department. *Pediatr Emerg Care* 30: 128-130. [Crossref]
10. Lui CT, Chan TW, Fung HT, Tang SY (2010) A retrospective study on imperforate hymen and hematometrocolpos in a regional hospital. *Hong Kong J Emerge Med* 17: 436-440.
11. Papes D, Arslani N, Rajkovic Z, Altarac S, Kopjar M (2011) An unusual cause of anuria and hydronephrosis in a 12-year-old girl. *Ren Fail* 33: 540-543. [Crossref]
12. Bakos O, Berglund L (1999) Imperforate hymen and ruptured hematosalpinx: a case report with a review of the literature. *J Adolesc Health* 24: 226-228. [Crossref]
13. Torres MK, Alensuela AB (2013) A case report on an unusual complication of imperforate hymen. *J Endometriosis*. Conference: 2nd European Congress on Endometriosis. Germany. 5: 54-55.