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Case Report

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Imperforate hymen mimicking hematocolpos: A case report and mini review of the literature

Abstract

Introduction: An imperforate hymen represents a rare congenital obstructive malformation of the vagina associated with hematocolpos (0.1% prevalence). The aim of this case report and mini literature review is to familiarize physicians with the condition and inform regarding prompt diagnosis and management.

Case report: Case of a 16-year-old patient with imperforate hymen complicated by hematocolpos. The patient initially presented at the outpatient clinic with cyclical pelvic pain, primary amenorrhea and a protruding fluid-filled hymen. After thorough evaluation, she underwent hymenotomy and drainage, without complications.

Discussion: A review of current literature regarding imperforate hymen was performed. The search words "imperforate" AND "hymen" were used.

Conclusion: Physicians should become familiar with the condition in order to diagnose and treat it in a timely fashion and with the appropriate technique, on an individualized basis. More evidence is needed regarding long-term outcomes post-hymenotomy/hymenectomy.

Keywords: imperforate hymen, hematocolpos, hematometrocolpos, hymenotomy, hymenectomy

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Introduction

An imperforate hymen depicts a rare congenital obstructive malformation of the vagina with a prevalence of 0.1%. It often presents with cyclical pelvic pain and primary amenorrhea at puberty. Due to its rarity, an imperforate hymen may not be diagnosed until serious complications have developed.[1–3] Aim of this case report and mini literature review is to familiarize physicians with the condition and encourage prompt diagnosis and appropriate management.

Case report

A 16-year-old female (G0, P0) was referred for evaluation at the outpatient gynecologic clinic of the tertiary hospital "Elena Venizelou" in Athens, Greece, due to primary amenorrhea and lower abdominal/pelvic pain that she had been experiencing for a year but had worsened in the last 3 months. Upon clinical examination, it was discovered that she had an imperforate hymen that presented as a protruding purple-bluish fluid-filled mass at the vaginal introitus (Figure 1).

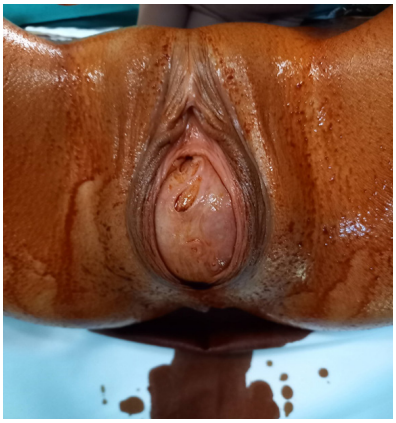

Figure 1 Fluid-filled hymen protruding at the intoitus.

She underwent an abdominal magnetic resonance imaging (MRI) scan, revealing a distended vagina measuring 18.4 x 8.8 x 8.2cm, with a T1 hyperintense and T2 hypointense fluid collection suggestive of hemorrhagic fluid, consistent with hematocolpos. The uterus measured 5.3x5.6x3.5cm and was displaced anteriorly and to the left of the midline (Figure 2) (Figure 3). The rest of the abdominal and pelvic organs, including the kidneys, appeared normal.

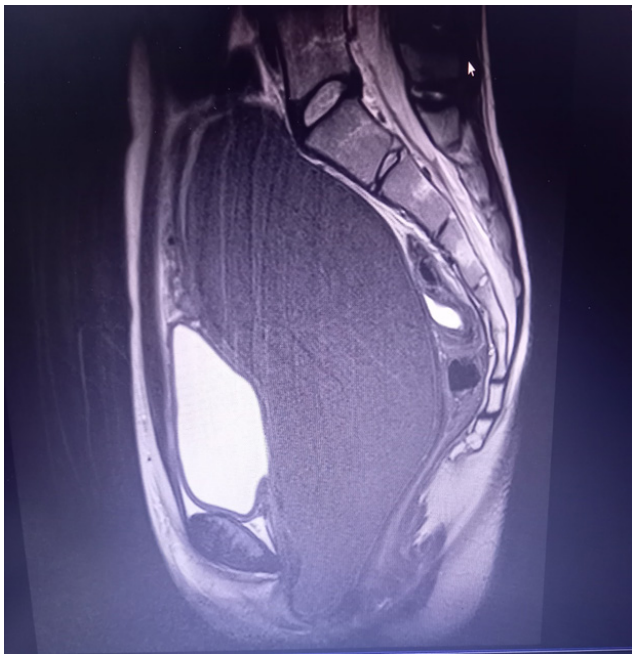

Figure 2 Presence of hematocolpos (MRI scan: sagital section).

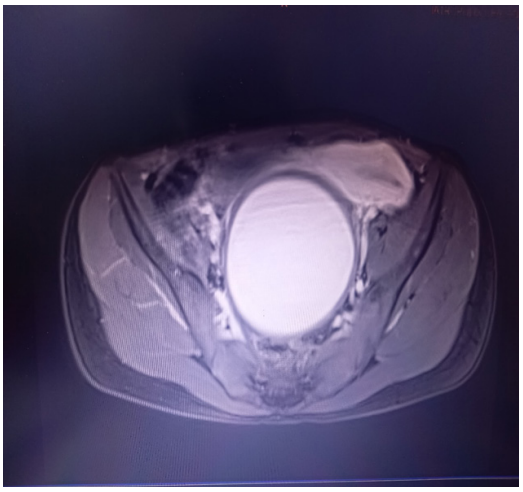

Figure 3 Presence of hematocolpos and displaced uterus (MRI scan: transverse section).

The patient underwent a hymenotomy and surgical drainage of the hematocolpos. A circular (annular) incision at the center of the bulging hymen was performed by electrocautery, followed by drainage of approximately 2500ml of thick brownish menstrual blood and diligent irrigation of the vagina (Figure 4). Care was taken to spare hymenal tissue circumferentially from the base in order to preserve virginity. A normal cervix was visualized and inspected. The vaginal entrance was surgically restored as follows: The edges of the vaginal margins were everted by suturing the inner vaginal mucosa to the exterior vestibular mucosa with absorbable No2 sutures, firstly by performing single interrupted sutures which were then followed by a purse-string suture (Figure 5). Vaginal swabs were collected and the results of the cultures were negative for infection. A penrose tube was placed at the introitus to ensure unimpeded continuous drainage and was removed on the second postoperative day. Broad-spectrum antibiotics were administered for post-operative chemoprophylaxis. The postoperative course was uncomplicated and the patient was discharged on the fourth postoperative day. Her wound had completely healed by the time she had her three-month post-op follow-up appointment (Figure 6). To this day, she has been experiencing normal painless menses.

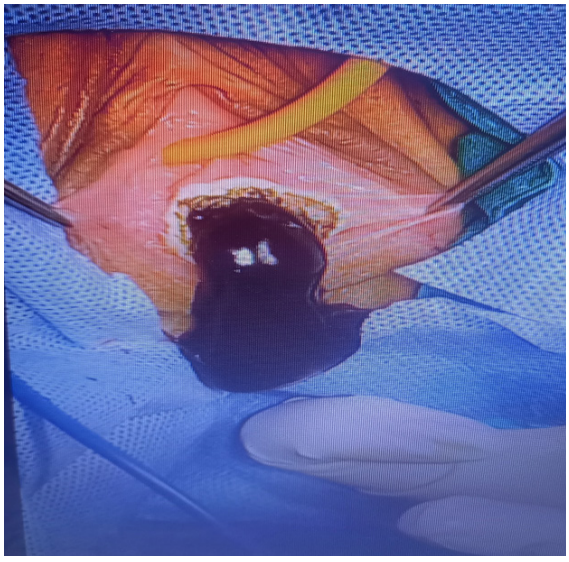

Figure 4 Surgical drainage of hematocolpos through an annular incision.

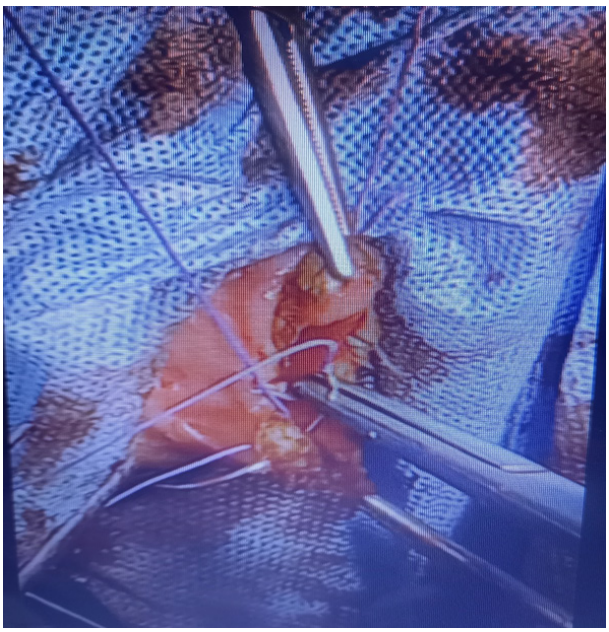

Figure 5 Placement of sutures at vaginal margins.

Figure 6 Fully healed introitus post-hymenotomy.

Discussion

An imperforate hymen depicts a rare congenital obstructive malformation of the vagina with a prevalence of 0.1% and an incidence of 1 out of 2000.[1,2,4] It was first described by French physician, Ambroise Paré in the 1500s.[3] Most cases are sporadic, although occurrence within families, with autosomal dominant or recessive mode of transmission, has been documented.[2] It has also been associated with syndromes such as McKusick-Kaufman syndrome and Bardet-Biedl syndrome, which present with polydactyly, congenital heart defects and genitourinary abnormalities.[5–7]

The hymen is a layer of connective tissue that is thought to originate from the urogenital sinus and which normally ruptures perinatally and remains as a thin perforate membrane at the level of the introitus,

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partially obstructing the vagina. The function of the hymen is unclear but is thought to serve as a physical barrier protecting from infections during the prepubertal period. Failure to rupture causes complete blockage of the vaginal canal, the condition known as imperforate hymen.[8,9]

It is common for patients with an imperforate hymen to be asymptomatic and undiagnosed until they reach the age of menarche.[10] However, in 30% of cases, neonates with the anomaly present with hydrocolpos (a distended vagina filled with fluid) or hydrometrocolpos (both vagina and uterus distended by fluid), caused by maternal estrogen and, thus, they are incidentally diagnosed with an imperforate hymen.[8,11] Prenatally, imperforate hymen may be associated with fetal ascites detected by ultrasonography.[4] The most common presentation of the entity, however, is primary amenorrhea with cyclical pelvic pain in puberty, as a result of the formation of hematocolpos or hematometrocolpos (that is a distension of the vagina or both the vagina and the uterus, due to the accumulation of obstructed menstrual blood).[1,8,10] The classic appearance is that of a bulging bluish hymen without a vaginal opening, as was the case with our patient. Urinary retention is also common (46-58% of cases), as well as abdominal acute pain, constipation and, rarely, intestinal obstruction.[2,4,9,12–14] Endometriosis due to retrograde menstruation has also been reported.[15]

Clinical findings and patient history are pivotal in diagnosing the condition. A transabdominal sonogram aids in detecting a distended vaginal canal in neonates and teenagers,[8,11] while a prenatal sonogram will detect fetal ascites caused by bladder outlet obstruction due to a hydrocolpos secondary to an imperforate hymen.[6] A computed tomography (CT) scan will also depict a distended fluid filled uterus and/or vagina, suggestive of hematometrocolpos and hematocolpos. The most important diagnostic tool, however, is a Magnetic Resonance Imaging (MRI) scan, which will depict both the distended vagina/uterus and protruding introitus. It will also assess the thickness of the imperforate hymen, as well as related complications such as infection, hydronephrosis, endometriosis. Kidney anatomy visualization is extremely important to exclude complex congenital syndromes.[4,11]

Differential diagnosis of an imperforate hymen should include the main causes of hemato/hydrocolpos; congenital anomalies and secondary vaginal obstruction due to infection, trauma or sexual abuse. The congenital causes that need to be excluded are distal vaginal agenesis, complete transverse vaginal septum and the Herlyn-Werner-Wunderlick syndrome (OHVIRA syndrome: obstructed hemivagina and ipsilateral renal anomaly).[11,16] Moreover, the differential diagnosis of the protruding interlabial mass of the hymen should include rare vaginal vascular malformations. Specifically, vascular venous malformations present as bluish soft compressible masses located at the vagina, while arteriovenous malformations might be pulsatile and are easier to bleed.[8] Finally, rare tumors of childhood, such as a rhabdomyosarcoma, should also be excluded. A rhabdomyosarcoma usually affects girls younger than six years of age and has the appearance of a grape-like cluster of prolapsing masses, which differs greatly from the soft bulge of a prolapsing hymen.[8]

The most common sequelae of an imperforate hymen complicated by a hematocolpos include infection (vaginitis, endometritis, salpingitis), urinary retention – hydronephrosis - acute bacterial nephritis - renal failure, tubal adhesions, pelvic endometriosis, infertility.[9,11,14]

Treatment of an imperforate hymen entails performing either a hymenotomy (surgical incision of the hymen) or a hymenectomy (surgical removal of the hymen), followed by drainage of the hematocolpos.[1,6,10,11,16–20] Choice of technique applied should be made with consideration to patients' wishes regarding virginity preservation. Various types of incisions have been reported when performing hymenotomy, the most common of which is a cruciate incision, which involves incising the hymen in a cross-shaped manner. Caparo's incision is a sagittal incision on the hymen which results in the formation of a "lipped" hymen and allows maintance of virginity. A simple elliptical, circular or annular incision is another technique of hymenotomy that preserves virginity. However, some authors[16] suggest that it presents an increased risk of vaginal stenosis and future dyspareunia. The Pozzi techinique consists of a transverse incision to the hymen and the constrictor muscles at 5 and 7 o'clock positions and suturing circularly at several points. However, according to some authors,[1] incisions at 5 and 7 o'clock positions should be avoided in order not to risk injury to Bartholin's glands. Graber's radial incision consists of a central hymenectomy followed by radial incisions of the hymenal ring. Less invasive techniques are available, including the use of carbon dioxide laser and the application of a Foley catheter which is placed at the center of the incision, in order to ensure drainage and avoid damage to the structure of the hymen.[1,6,10,11,16–20] It should be noted that if a hematosalpinx is suspected, a laparoscopy should be performed prior to the hymenal incision in order to ensure proper drainage of the adnexa. Moreover, it is suggested that suprapubic pressure should not be performed during drainage for faster evacuation, because it may result in blood reflux that may cause pelvic endometriosis. Broad-spectrum antibiotics should always be given for post-surgical prophylaxis.[1,16]

Complications of hymenotomy and hymenectomy include bleeding, vaginal and pelvic infection, scarring, vaginal adhesions and stenosis and are reported in approxiamately 6.6% of cases. Placing sutures (regardless of method) around the re-created orifice seems to prevent re-fusion.[17] It is worth noting that the risk of hymen re-fusion is believed to be higher in prepubertal patients because their genital tissue lacks estrogen.[21] According to literature available, there are no differences in outcomes among methods. However, some authors[10] have reported that hymenectomy is associated with more frequent complications (i.e. bleeding) than hymenotomy, such as bleeding, while others state that the less invasive techniques present a higher risk for recurrence.[14] Regardless of technique choice, care should always be taken to avoid injuring the urethra and Bartholin's glands. The optimal timing of the operation depends on symptoms. In general, if a child is asymptomatic, it should undergo surgery at menarche, before the development of hematocolpos or hematometra. Symptomatic children should, of course, receive immediate treatment.[6]

Due to the rarity of an imperforate hymen, there is lack of evidence regarding long term gynecologic and obstetric outcomes, although most authors believe them to be good as long as the patient does not develop major complications, either iatrogenic (post-surgery) or primary due to delayed diagnosis (i.e. pelvic infection or endometriosis).[6,11,14] There is, however, a retrospective study conducted by Amitai et al.,[22] according to which women who had undergone hymenectomy for imperforate hymen exhibited increased rates of dyspareunia, endometriosis, infertility, cesarean deliveries (due to either malformation of the birth canal or bias of obstetricians against vaginal labor when patients have a history of a hymenotomy) and vaginal and/or perineal lacerations during vaginal labor. The afore-mentioned study, however, has several limitations, the most important of which is the small sample size of 56 patients as a result of the rarity of the condition.[22]

Conclusion

In conclusion, an imperforate hymen is a rare congenital condition which, if left untreated, may lead to serious complications both acute and long-term for the patient. Therefore, pediatricians, neonatologists and gynecologists should be familiar with the condition in order to diagnose it sooner rather than later, ideally at birth. Emergency medicine physicians should also be aware of the condition and suspect it when pubertal patients present at the emergency department with acute pelvic pain and a history of primary amenorrhea. Early detection and appropriate management is paramount in order to prevent serious complications of an imperforate hymen.

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Conflicts of interest

All authors declare no conflicts of interest with respect to this manuscript.

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