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Case Report

Hematocolpos Secondary to an Unrecognized Diagnosis of Hymenial Imperforation: A Case Report and Review of the Literature

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Abstract

A relatively rare congenital malformation, hematocolpos is the progressive accumulation of menstrual blood in the vaginal cavity at puberty. It is often the consequence of a hymen imperforation. It is clinically manifested by cyclic pelvic pain and primary amenorrhea and, more rarely, by a pelvic mass syndrome. The diagnosis is primarily clinical. Ultrasound and magnetic resonance imaging (MRI) are additional tests to confirm hematolcolpos and exclude other associated genitourinary malformations. Treatment consists of a hymenotomy to drain the hematocolpos. The age of discovery of hematocolpos varies from 13 to 14 years. We report a case of hematocolpos secondary to hymenal imperforation diagnosed in a 17-year-old girl with periodic pelvic pain, primary amenorrhea and a pelvic mass. A first ultrasound trap had evoked a large ovarian cyst but the repeat pelvic ultrasound in our hospital confirmed the diagnosis of hematocolpos.

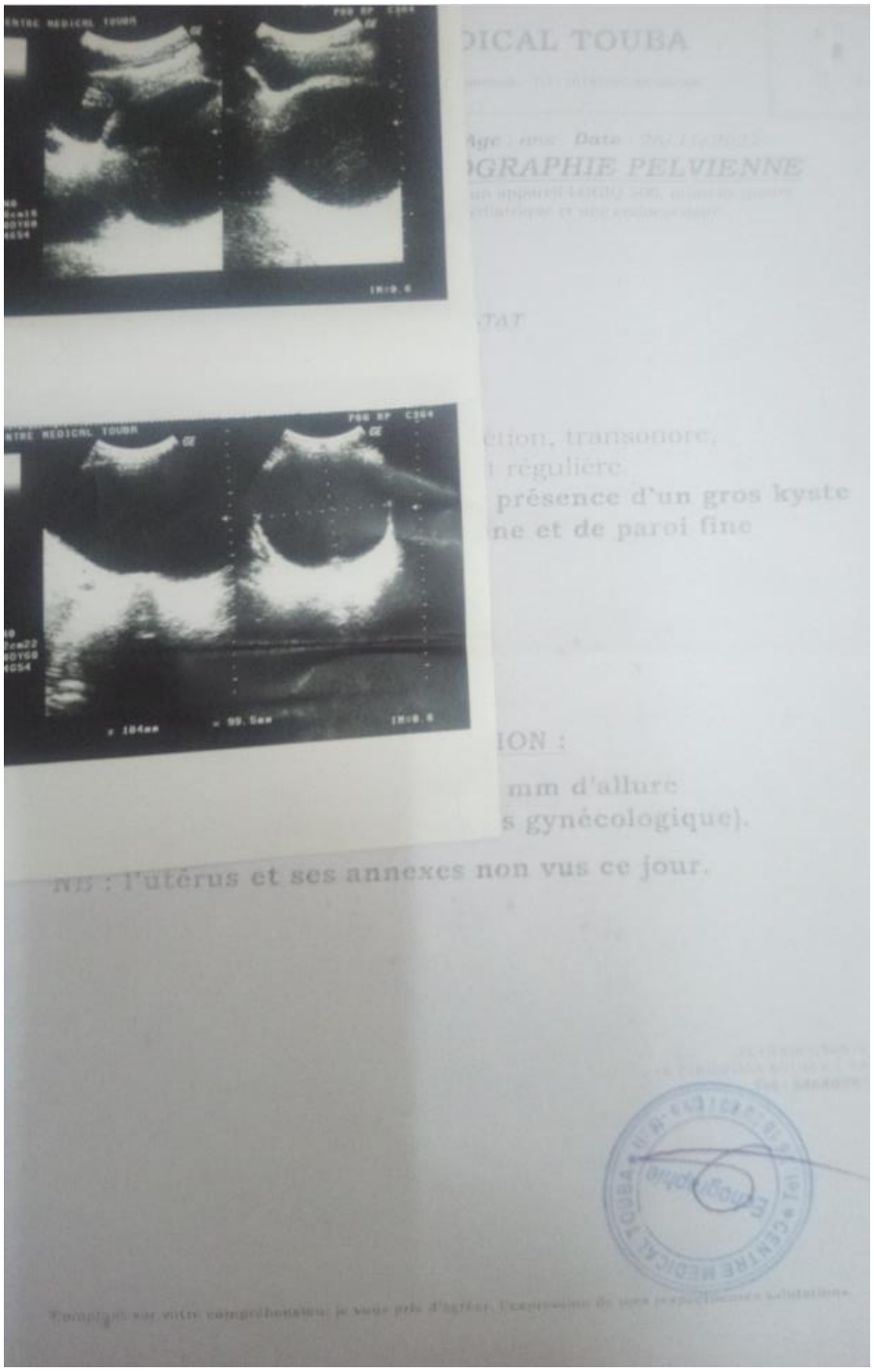
Keywords: Hymenal imperforation, hematocolpos, cyclic pain, amenorrhea, hymenotomy.

INTRODUCTION

Hymenal imperforation is a relatively rare condition, but the most common congenital malformation of the female genital tract. It was first described by Ambroise Paré in 1633 [1, 2]. It causes an obstruction of the vulvar orifice responsible for the vaginal retention of menstrual blood at puberty which is called: "hematocolpos". Hymeneal imperforation is often isolated, its diagnosis which seems easy from birth by systematic screening, is usually made at puberty by inspection of the external genitalia [1-4]. Painful cryptomenorrhea is the most frequent symptom. Non- gynecologic symptoms can sometimes mislead the diagnosis [1-5]. Early diagnosis and treatment of hymenal imperforation is important to avoid tubal sequelae. Ultrasound is the examination of choice for the diagnosis of hematocolpos on hymenal imperforation. Magnetic resonance imaging (MRI) is the reference imaging test to confirm hematocolpos and exclude other Muller's canal malformations or associated urological malformations [6-8]. Treatment is exclusively surgical: "Hymenotomy. Systematic screening at birth and early treatment are the best guarantees for the prevention of complications of this pathology [16, 17]. We illustrate our remarks with a clinical case reporting the positive diagnosis of hematocolpos by hymenal imperforation in a 17 year old girl presenting with primary amenorrhea and a pelvic mass and who was hospitalized and treated in the gynecology department of the Mali hospital in Bamako.

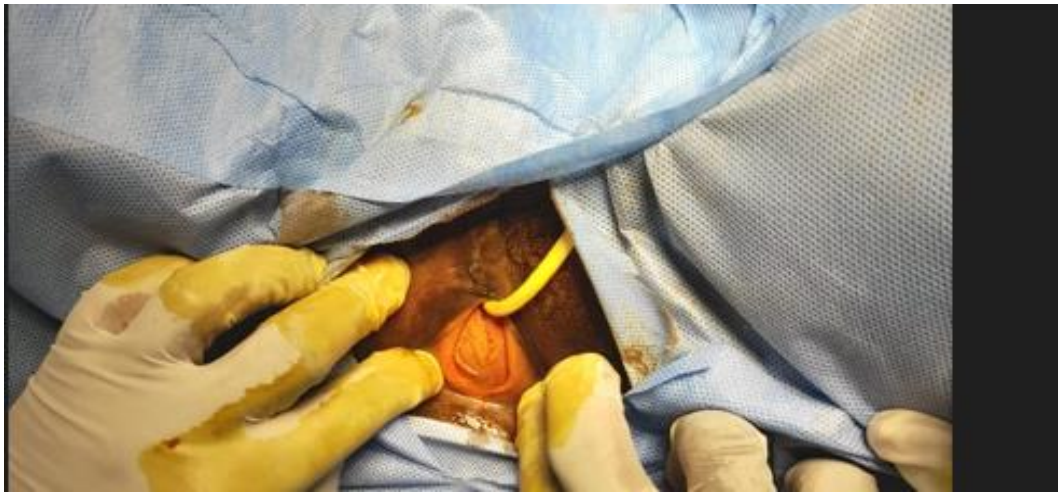
CLINICAL CASE: PATIENT AND OBSERVATION

A 17 year old girl accompanied by her father consulted the gynecology department of the hospital for periodic pelvic pain contemporaneous with menstruation and evolving for several years. It was a very intense cyclic pelvic pain with heaviness radiating in the lumbar fossa and the back. The associated signs were the absence of menarche and the sensation of a pelvic mass. The patient had in her position the report of a previous pelvic ultrasound performed in a local health facility evoking a large pelvic cyst of 104X99 mm of functional appearance (Interest of a gynecological opinion). NB The uterus and adnexa not seen today).

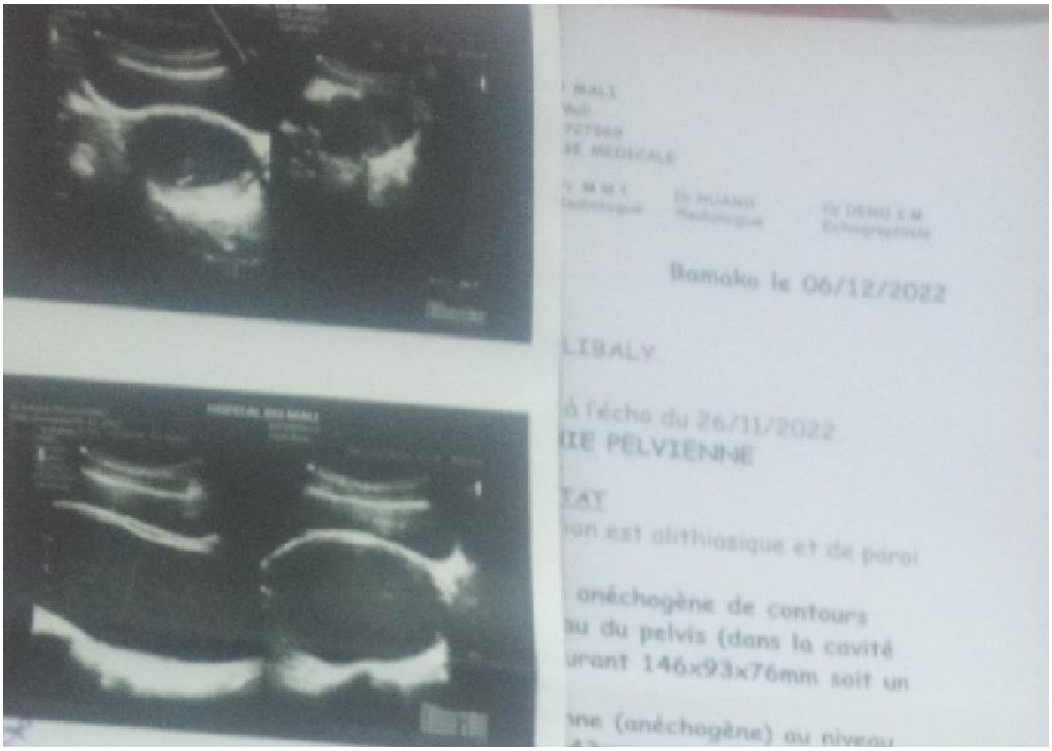


On clinical examination, hemodynamic and staturo-weight parameters were normal. Pubertal development was of adult type according to Tanner's classification scale (breast development: Stage 5: breasts reaching their adult anatomy; pubic hair: Stage 5: hair reaching adult type and extending over the inner

surface of the thighs). Abdominal examination revealed a firm renal pelvic mass with sub-umbilical pain on palpation. Pelvic examination showed a bulging, imperforate hymen.

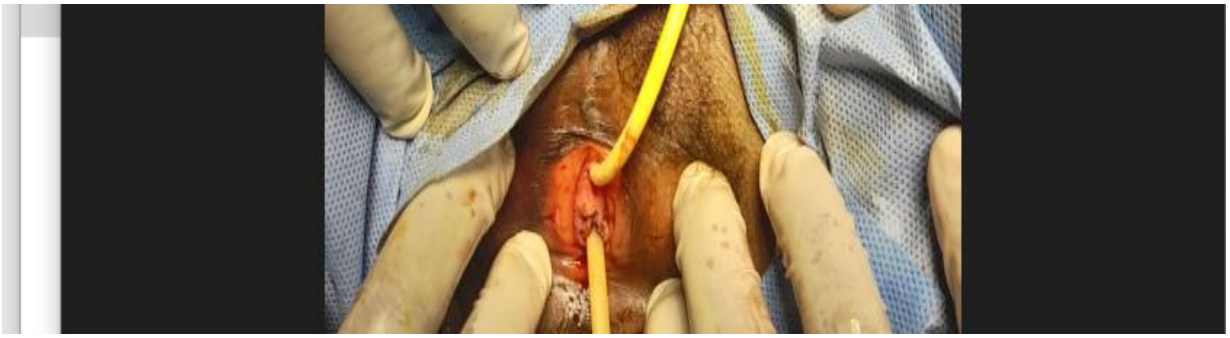


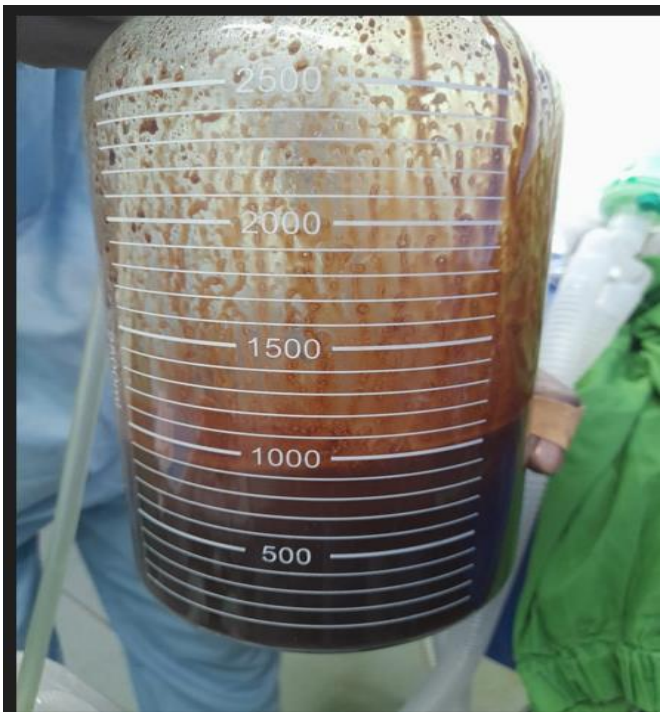
The rest of the examination was unremarkable. We concluded that she had a hymenal imperforation associated with chronic cyclic pelvic pain and primary amenorrhea. As a check-up, we asked for a repeat pelvic ultrasound at the imaging department of the Mali hospital, which revealed an anechoic mass with regular contours, well limited at the pelvic level (in the uterine cavity) in the retrovesical area, measuring 146 x 93 x 76 mm, i.e. a volume of 520 ml.mm, and also noted the presence of a liquid image (anechoic) at the left tubal level measuring 57 x 43 mm. The conclusion of this ultrasound was: ultrasound aspect in favor of hematocolpos of great abundance associated with the left hematosalpinx.



We did not request an MRI because of the low purchasing power of the patient's parents, who were barely able to afford the standard preoperative biological workup. The patient was taken to the operating room under general anesthesia. We performed the hymenotomy by cruciform incision bringing back about 1000 cc(ml) of black-looking hematic fluid. A foley catheter ch16 inflated to 10CC was left in place through this vaginal orifice for 05 days before its removal. The bladder catheter was removed 24 hours after the procedure. The virginity was preserved by our type of very minimal incision. The postoperative course was simple and the patient was discharged on the 5th day after the operation. Antibiotic coverage was

prescribed with Rapiclav 1g morning and evening for 7 days.





RESULTS AND DISCUSSION

The hymen is derived from the interface between the urogenital sinus and the ducts of Muller which fuse to form the uterus. It is composed of two epithelial blades interposed by connective tissue which partially degenerates during the fifth month in utero, making the vaginal lumen communicate with the vestibule. The hymenal imperforation could be due to a defect of apoptosis or to an inappropriate hormonal environment. The hymen is a remnant of the mesodermal sheet that normally perforates during the later stages of embryonic development [1-3]. Hymenal imperforation is a rare event estimated at 1 in 2000 female births [1, 3]. The incidence reported in the literature is widely variable, depending on whether it is assessed globally, by age or by the type of anatomical lesion [3]. In typical cases, the age of discovery of hematocolpos is between 12 and 15 years (the age of menarche) [3]. In our clinical case the patient was 17 years old (period of first post-pubertal sexual intercourse). The prevalence of hymenal imperforation is 0.1% [1, 3]. Familial character is exceptional. The majority of cases reported in the literature are sporadic, however, a few familial cases have been described suggesting a probable genetic predisposition [1, 3]. Our patient had no family history of hymenal imperforation. The diagnosis of hymenal imperforation is possible in utero when a hydrometrocolpos is found on ultrasound [3]. In utero diagnosis has the added advantage of looking for associated renal malformations. This diagnosis can be made by systematic screening at birth but also in the presence of hydrometrocolpos during the genital crisis of the female newborn [3]. Most often, this malformation is discovered at puberty. Secondary sexual characteristics are present, contrasting with primary amenorrhea. The typical clinical picture is dominated by cyclic median pelvic pain that may sometimes take on a pseudo-appendicular character, sometimes accompanied by lumbosciatalgia. A pelvic mass syndrome may, in case of a large hematometria, cause urological and/or digestive complications such as urinary retention, dysuria, hydronephrosis or

constipation. The cyclical nature of the painful attacks may be missed due to the usual irregularity of the menstrual cycle during the peripubertal period. The pain may be deceptive, pseudo appendicular and may lead to 'excessive' interventions for suspicion of acute appendicitis. The diagnosis of hematocolpos is clinical. It is made during the gynecological examination; on inspection of the vestibule, an intact, semi-transparent, bulging, bluish membrane is observed [3-10]. Our patient had good external genitalia development, and secondary sexual characteristics. She also had cyclic pelvic pain previously with primary amenorrhea. Blood is retained first in the vagina, then the uterus (hematometry) and eventually the fallopian tubes. Its volume varies from patient to patient and can even reach 3 liters [3, 9, 10]. Retrograde menstrual flow may alter the fallopian tubes or lead to endometriosis lesions that may interfere with subsequent fertility. However, this is rare if the diagnosis is made early and fertility is usually preserved [3, 9, 10]. In one case, the genital examination showed the existence of a hymenal imperforation with a bulging imperforate hymen in the patient, and palpation showed the existence of a pelvic mass [3, 9, 10]. [3, 9, 10]. Pelvic ultrasound and nuclear magnetic resonance, because of their safety in adolescents, are not only useful for confirming the diagnosis in case of doubt, but also for demonstrating the presence of a mass in the pelvis.

Of doubt, but also to detect any malformations or associated complications [3, 10]. The malformations to be systematically investigated when a hymenal imperforation is discovered are genital (utero-vaginal duplication), nephro- urinary (urinary malformations, renal agenesis, dysplasia or hypoplasia) and anorectal. Complications associated with hematocolpos are infections (endometritis, salpingitis, tuboovarian abscess) and pelvic endometriosis favored by retrograde menstruation. These complications have an impact on the future fertility of the adolescent [3-10]. In this case, the first ultrasound scan had mistakenly suggested a large pelvic cyst, but the abdominopelvic ultrasound scan performed in our hospital revealed a uterine and intra-vaginal fluid collection in favor of a hematocolpos. The treatment was surgical. The objectives of this treatment are: ? Restore the permeability of the genital tract? To ensure normal sexual function? To try to preserve future fertility. It must be undertaken in all cases, one should not count on the spontaneous regression of retentions. Abstention, even in mild forms, would risk allowing genital and urinary infections to develop, which are more frequently encountered in forms detected late. In many cases, treatment is limited to simple drainage of the retained pouch. The hymenotomy must allow a normal menstrual flow, trying to respect as much as possible the virginity of these young patients, especially in our social context, and to ensure a normal sexual life afterwards. The treatment of hymenal imperforation is, above all, surgical. It consists of a hymenotomy. Its aim is to drain the hematocolpos and restore vaginal flow. Several incisions have been described: vertical, T-shaped, cross, radial and circumferential incisions [11, 16, 17]. Circumferential incisions should be avoided because they lead to orifice stenosis, a source of dyspareunia. The hymenotomy must meet two requirements: to respect the orifices of Bartholin's glands and to encourage urethro-hymenial disassociation. In addition, it is recommended not to exert uterine pressure during emptying so as not to encourage tubal reflux responsible for pelvic endometriosis. Antibiotic therapy is prescribed during the operation to avoid any infectious complications. In all cases, a postoperative clinical check-up must be systematic to verify the absence of secondary stenosis [11, 16, 17]. This said, the best treatment remains preventive, based on an early diagnosis of the malformation and on surgery undertaken after the development of the genitalia but before the appearance of hematocolpos. In our case the patient had a cruciform incision (Cross), evacuation of the hematic collection, and a drainage by a probe left in place for 5 days was principle. The patient was put on broad-spectrum antibiotics and the catheter was removed on the 5th day after the operation, with protection worn for 3 to 4 days. The evolution was satisfactory without postoperative stenosis.

CONCLUSION

Hymenal imperforations are often benign conditions, with a favorable evolution, if diagnosed and treated early. On the other hand, if it is not recognized, it can lead to serious complications, threatening the vital prognosis and seriously compromising the obstetrical future. Systematic screening at birth and early treatment are the best guarantees of prevention of complications of this pathology.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

Contribution of the Authors

The presentation of the clinical case was adopted by Seydou Mariko, Brahima Bamba and Amaguiré Saye. The drafting of the manuscript was carried out by Seydou Mariko and Brema Bamba, Amaguiré Saye had supervised the elaboration of the manuscript until its submission. All authors had participated substantially in the development of the manuscript and were willing to take public responsibility for the work. All authors have seen and approved the manuscript as submitted.

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