

## CASE REPORT

# SPONTANEOUS OPENING OF IMPERFORATE HYMEN IN AN ASYMPTOMATIC FIVE YEAR OLD NIGERIAN GIRL

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### ABSTRACT

**Aim:** To present a rare case of spontaneous rupture of asymptomatic imperforate hymen.

**Case report:** Spontaneous rupture of the hymen has been reported in neonates and adolescent girls with symptomatic imperforate hymen (when neonates present with hydrocolpos, hydrometrocolpos, or mucocolpos; and adolescent girls present with haematocolpos, and hematometra), but to the best knowledge of the researcher, no case of spontaneous rupture of imperforate hymen has been reported in asymptomatic preadolescent girls. We present a 5 year old girl who had spontaneous rupture of asymptomatic imperforate hymen.

**Conclusion:** The occurrence of spontaneous rupture of hymen should be considered not only in neonates and adolescents with symptomatic imperforate hymen, but also in asymptomatic young girls.

**Key words:** Imperforate hymen, spontaneous rupture, asymptomatic

### INTRODUCTION

Imperforate hymen (IH), is a congenital anomaly, in which the hymen has no opening and occludes the vagina opening.<sup>1</sup> Although, it is the most common and most distal form of vaginal outflow obstruction, it is a rare congenital anomaly in girls with an incidence of 0.014-0.1%.<sup>2-4</sup>

During the 12<sup>th</sup> week of gestation, the lower part of the vagina is formed from the canalization of the vaginal plate. This process is completed by the fifth month of gestation.<sup>5-7</sup> The hymen is formed from the proliferation of the sinovaginal bulbs or vaginal plates.<sup>6,8</sup> The central portion of this membrane perforates due to the degeneration of its epithelial cells to form the vaginal orifice/perforated hymen. Incomplete canalization of the hymen results in an

imperforate, microperforate and septate hymen.<sup>9</sup>

The configuration of the hymen varies from child to child and changes as the newborn grows and matures into adolescence.<sup>9</sup> The three most common configurations are annular, crescentic, and fimbriated.<sup>9</sup> The annular hymen has a doughnut like appearance; a crescentic hymen has no definable hymen tissue between the 11 and 1 O'clock positions anteriorly; while the fimbriated hymen forms redundant projections, which overlap and obscure the hymenal orifice. Another variant is the sleeve-like hymen, which is redundant and thickened. Both the fimbriated hymen and sleeve-like hymen are seen typically in infants with residual maternal estrogen. As they mature and estrogen levels drop, the hymen becomes more annular, and at age 4 to 9 years the hymen becomes more crescentic.<sup>1,9</sup> Other hymenal configurations include septate and cribriform hymen. The former, usually resolves as

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the child develops or ruptures spontaneously. A cribiform (sieve-like) hymen is defined by multiple openings in the hymen.<sup>1,9</sup> A normal hymen with adherent edges (similar to cribiform hymen) has been described in literature.

Based on the mode of clinical presentation, IH can be classified as either symptomatic or asymptomatic. Diagnosis of asymptomatic imperforate hymen is most commonly made during routine newborn examination of the female genitalia, while symptomatic is made most commonly in the adolescent with symptoms of vaginal outflow obstruction and primary amenorrhoea.<sup>2,4,10,11</sup>

Only few cases of symptomatic imperforate hymen have been reported in infancy and premenarchal girls.<sup>3,12,13</sup> In a case report series, involving 20 cases of IH, only one was a 2 year old girl, while the others were all adolescent girls.<sup>12</sup> A rare case of IH associated with bicornuate uterus as a cause of bilateral hydronephrosis and pelvic mass in an 8 month old infant was reported in Turkey in 2012.<sup>11</sup> A similar report was made in Japan, in a 35 day old infant who presented with features of acute renal failure; abdominopelvic imaging revealed bilateral hydronephrosis and hydrometrocolpos.<sup>3</sup> In Nigeria, a few studies have reported the occurrence of imperforate hymen in neonates and infants. In Northern Nigeria, 2 cases of symptomatic IH was reported in infants who presented with hydrometrocolpos.<sup>14</sup> In Southern Nigeria, in a case report series, a diagnosis of IH was eventually made after being missed initially in 3 neonates who presented with hydrometrocolpos.<sup>15</sup>

Clinical presentation of symptomatic IH include: abdominal mass, bulging hymen, bilateral hydronephrosis, and features of urinary retention.<sup>3,12-15</sup> All of these symptoms resulted from the formation of hydrometrocolpos, which is a tense and gray-white appearing vestibule behind the IH.<sup>2,4,11,16,17</sup> It is due to the persistent effect of maternal estrogen in the vagina, which then leads to excessive accumulation of vaginal and cervical secretions behind the hymen. However, most infants with IH are asymptomatic

because the accumulation of vaginal and cervical secretions is usually small and resolves with time following the cessation of the estradiol stimulation.<sup>18</sup> Thus, these infants may go unnoticed until they become symptomatic during adolescence, when they attain menarche.<sup>18</sup> This may explain why it is rare to make a diagnosis of asymptomatic IH outside of the neonatal period and before menarche. In Nigeria, none has been reported.

Hymenotomy or Hymenectomy is the surgical intervention often required in the adolescent, infant, and young child who presents with symptomatic vaginal outflow obstruction.<sup>3,19-21</sup> However, in asymptomatic infants and young girls, intervention is delayed until they approach menarche so as to allow adequate amount of estrogen, which helps with healing and may counter the need for reoperation.<sup>12,18</sup> Spontaneous rupture of the hymen could occur in symptomatic neonates and adolescents, but none has been reported in asymptomatic young children.<sup>22,23</sup>

In this report, we showcase not only the rare occurrence of IH in a 5 year old, but also the spontaneous opening of asymptomatic imperforate hymen. To the best knowledge of the author, spontaneous rupture of IH has been reported in symptomatic children, but none in an asymptomatic 5 year old girl.

## CASE REPORT

We report CE, a 5 year old girl, who presented to the Children Outpatient Department accompanied by her mother, who is a 40 year old Business woman, with tertiary level of education. The presenting complaint was absent vagina opening, which mother claimed she noticed one week prior to presentation. The mother noticed that the vagina of her 5 year old niece was different and looked normal, compared to that of her child. She claims she did not notice this earlier. CE resides with her parents in Port

Harcourt, southern Nigeria; she only came along with her mother to Benin City, for the Holiday.

On Examination, she was calm and co-operative, in no obvious distress. In lithotomy position, the Labia majora and minora were gently parted, the Vulva was seen, the urethra orifice was visualised and intact but the hymenal/vaginal orifice was absent. A thin groove measuring about 1cm long on the hymen just below the urethral orifice was seen. A diagnosis of imperforate hymen was made (Figure 1).

A Pelvic Ultrasound Scan, showed normal female reproductive organs including the uterus, which measured 2.93cm X 0.66cm (Figure 2). Following review by the Gynaecologist, it was observed that the groove on the hymen (noticed on first examination), deepened a bit following parting of the labia, for proper visualization of the hymen. The gynaecologist, confirmed the diagnosis of imperforate Hymen (Figure 3).

The mother was counseled on the need for hymenectomy at the onset of puberty. She was advised to present at the hospital through the OPD, at the onset of puberty, marked by onset of breast development. However, 24hrs later, she presented to the COPD with reports of spontaneous opening of the vaginal orifice (Figure 4). Mother claimed no attempt was made at home by her or any other adult to forcefully and locally perform hymenotomy, no history of bleeding per vagina. Mother claimed she checks the hymen daily, expecting it to miraculously open. On examination of the perineum, it was pink, no evidence of trauma, no bruises, nor swelling; no bleeding, no tenderness, the vaginal orifice was present and patent; it was well rounded with smooth edges. She was asked to return for follow up one week later. During follow up visit, vaginal opening was still patent; patient's mother was counseled to bring her for yearly checkup, if there will be need for a further surgery especially when she attains menarche and coitarche. This is important to prevent clinical conditions such as hydrometrocolpos, and vaginal tears.



Figure 1: Groove seen on the hymen during first examination by the paediatrician

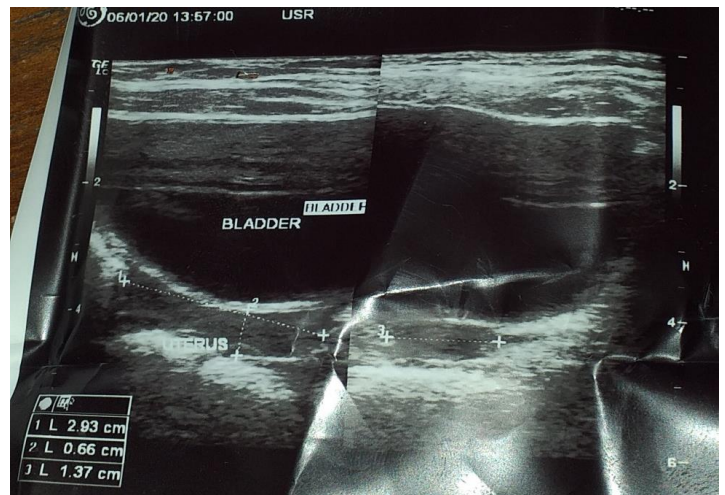
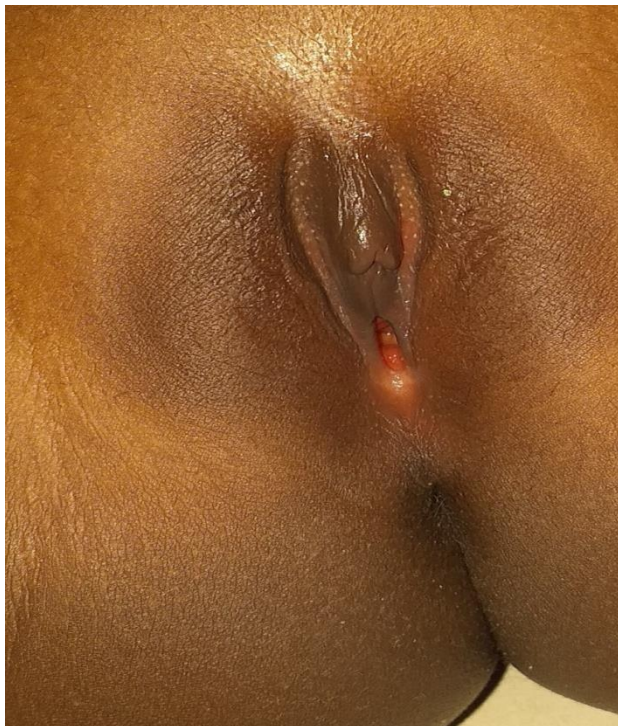


Figure 2: Abdominopelvic Ultrasonography, showing infantile uterus, patent vagina.



**Figure 3: groove more obvious during second examination by the Obstetrician**



**Figure 4: Spontaneous perforation, showing perforated hymen with smooth edges.**

## DISCUSSION

In utero, imperforate hymen is diagnosed during obstetric ultrasonography, when a foetus develops hydrocolpos, hydrometrocolpos, or mucocolpos.<sup>24,25</sup> In the neonates, asymptomatic IH is most commonly diagnosed during routine newborn examination; however, most midwives and paediatricians find this challenging due to variations in hymenal anatomy. This may explain why in this index case, the diagnosis was missed in infancy. In addition, it is possible that the hymen was not examined at birth, because of a low index of suspicion; and also, thorough routine newborn examination of the hymen is not a common practice in most health care facilities in Nigeria.<sup>15</sup>

From post neonatal period to prepubertal period/age, diagnosis of imperforate hymen is rare, except in some children who present with features and complications of hydrometrocolpos, such as urinary tract obstruction.<sup>3</sup> Most cases at this age are asymptomatic and discovered incidentally as seen in this case report. Thus, it is recommended that examination of the hymen should be done at least once in childhood, during visits to the paediatric clinics and findings documented.<sup>13</sup>

Adolescence is the most common period when diagnosis is made; they present with primary amenorrhoea, cyclical abdominal pain, abdominal mass, haematocolpos, and hematometra.<sup>26</sup> Late diagnosis has been associated with increased morbidity (including the problem of pelvic adhesions, intra-abdominal endometriosis, and urinary outlet obstruction) compared to when diagnosis is made early, prior to onset of symptoms and complications of vaginal outflow obstruction.<sup>11,26</sup>

Definitive treatment of IH is surgical.<sup>26,27</sup> Surgical intervention is usually reserved for children with symptomatic IH, and for asymptomatic pre-menarchal pubertal girls after the onset of oestrogen production signaled by onset of breast development.<sup>22,27</sup> This is because healing is optimal, risk of scarring is reduced and the need

for reoperation is avoided by oestrogen production.

Hymenotomy and hymenectomy are the two basic types of surgical intervention.<sup>27,28</sup> The classical type is the hymenectomy, which is a cruciate incision made on the hymen and the edges are incised to prevent closure, while hymenotomy is a simple vertical incision on the hymen to allow for the free flow of menstrum, the orifice is then enlarged by making a circular incision/annular hymenotomy.<sup>4,27</sup> The latter procedure seems less invasive but may have the risk of re-closure. It is considered a hymen sparing surgical intervention and in a systematic review, there was no difference of improved outcome between hymenotomy and hymenectomy.<sup>27</sup> In cultures and religion, such as seen in Nigeria, where people emphasize on virginity, hymen sparing procedure is readily preferred.<sup>4,27</sup> Other hymen sparing techniques include: using a simple vertical incision with oblique sutures to prevent refusion,<sup>20</sup> making a small central incision with the placement of a foley catheter, which is then left in place for 2 weeks, accompanied by the application of estrogen cream.<sup>20</sup> These are less invasive and preferable by most patients and families.<sup>19-21</sup>

Spontaneous rupture of IH is rare, but can occur.<sup>22,23</sup> A few cases have been reported in literature.<sup>22,23</sup> Spontaneous rupture of hymen has been reported in 2 newborns with mucocolpos;<sup>22</sup> also, a 13 year old adolescent with haematocolpometra had spontaneous rupture of membrane just before a planned hymenotomy operation.<sup>23</sup> This phenomenon of spontaneous rupture of IH occurred due to the increased intravaginal pressure from the haematocolpometra as seen in the 13 year old or mucocolpos as seen in the newborn babies. However, this cannot explain the occurrence of spontaneous rupture of IH in this index case we reported.

To the best knowledge of the authors, no literature has reported the occurrence of spontaneous rupture of IH in asymptomatic girls. This is the first. In this index case report, there

were no symptoms of vagina outflow obstruction, no intravaginal collection and thus no intravaginal pressure acting on the hymen to cause a rupture. The groove on the hymen which was noticed on first examination may have been a point of weakness, and due to repeated pressure on the hymen (including repeated physical examination) led to spontaneous rupture of the hymen. Considering the fact that there was no evidence of trauma to the vagina, no bleeding nor any sort of forceful manipulation as shown by the well circular opening and smooth edges (unlike that seen in adolescent girls with spontaneous rupture of IH, where deflorations are usually demonstrated),<sup>23</sup> the probability of a locally performed hymenotomy is low. Although it may be inferred that the groove in the hymen in this child may represent an atypical form of imperforate hymen or a normal variant of hymenal configuration, which spontaneously ruptured following mild perineal pressure or manipulation, no such case has been reported.<sup>1,9</sup>

In conclusion, this case report has exposed the need to consider the possibility of spontaneous opening of hymen occurring in girls with asymptomatic IH. Therefore, we recommend that the possibility of non-surgical intervention such as non-invasive progressive perineal manipulation could be the first line of treatment in cases of asymptomatic imperforate hymen.

## REFERENCES

1. Wallace NG, Amaya M. Normal and Developmental variations in the Anogenital examination of children. In: Jenny Carole, editor. Child abuse and neglect: Diagnosis, treatment and evidence. USA, Elsevier (Saunders). 2011. pp. 69-72
2. Nagai K, Murakami Y, Nagatani K, Nakahashi N, Hayashi M, Higaki T, Ishii E. Life threatening acute renal failure due to imperforate hymen in an infant. Paediatrics International. 2012;54:280-282

3. Eksioglu AS, Maden HA, Cinar G, Yildiz YT. Imperforate hymen causing bilateral hydronephrosis in an infant with bicornuate uterus. *Urology Case Reports*. 2012;102683. doi: 10.1155/2012/102683.
4. Basaran M, Usal D, Aydemir C. Hymen sparing surgery for imperforate hymen: case reports and review of literature. *J Pediatr Adolesc Gynecol*. 2009;22:61-64. doi: 10.1016/j.jpog.2008.03.009.
5. khemchandani S, Devra A, Gupta S. An unusual case of urinary tract obstruction due to imperforate hymen in an 11 month old infant. *Indian J Urol*.2007;23:298-299;
6. Shaw LM, Jones WA, Brereton RJ. Imperforate hymen and vaginal atresia and other associated anomalies. *J R Soc Med*. 1983;76:560-566
7. Acien P, Acien MI. The history of female genital tract malformation classifications and proposal of an updated system. *Hum Reprod Update*. 2011;17:693-705
8. Kuritan T. Development origin of vaginal epithelium. *Differentiation*. 2010;80:99-105
9. Smith A. The prepubertal hymen. *Australian Family Physician*. 2011; 40:873-875
10. Dane C, Dane B, Erginbas M, Cetin A. Imperforate hymen-a rare cause of abdominal pain: two cases and review of the literature. *J Pediatr Adolesc Gynecol*. 2007; 20:245-247.
11. Ercan CM, Karasahin KE, Alanbay I, Ulubay M, Baser I. Imperforate hymen causing hematocolpos and acute urinary retention in an adolescent girl. *Taiwanese Journal of Obstetrics and Gynecology*. 2011; 50:118-120.
12. Doyle JC. Imperforate Hymen: with and without Hematocolpos. A review of the literature and a report of twenty cases. *California and Western Medicine*.1942;56:242-247
13. Jill C. Posner and Philip R. Spandorfer. Early Detection of Imperforate Hymen Prevents Morbidity From Delays in Diagnosis. *Pediatrics*.2005;115:1008-1012.
14. Ameh EA, Mshelbwala PM, Ameh N. Congenital vaginal obstruction in neonates and infants: Recognition and management. *J Pediatr Adolesc Gynecol* 2011;24:74-78.
15. Okoro PE, Obiorah C, Enyindah CE. Experience with neonatal hydrometrocolpos in the Niger Delta area of Nigeria: Upsurge or increased recognition? *Afr J Paediatr Surg* 2016;13:161-165
16. Khan ZA, Rajesh U, Rastogi P, Joels LA. Imperforate hymen: a rare case of secondary amenorrhea. *J Obstet Gynaecol*. 2011;31: 91-92.
17. Abu-Ghanem S, Novoa R, Kaneti J, Rosenberg E. Recurrent urinary retention due to imperforate hymen after hymenotomy failure: a rare case. *Urology*. 2010;78:180-182
18. Finkel MA. Technical conduct of the child sexual abuse medical examination. *Child Abuse & Neglect*. 1998;22:555–566.
19. Cetin C, Soysal C, Khatib G, Urunsak IF, Cetin T. Annular hymenotomy for imperforate hymen. *J. Obstet. Gynaecol. Res*. 2016;42:1013–1015.
20. Ali A, Cetin C, Nedim C, Kazim G, Cemalettin A. Treatment of imperforate hymen by application of Foley catheter. *Eur. J. Obstet. Gynecol. Reprod. Biol*. 2003;106:3–4.
21. Friedman, M, Gac D, Peretz B. Management of imperforate hymen with the carbon dioxide laser. *Obstet. Gynecol*. 1989;74:270–272.
22. Kurdoglu Z, Kurdoglu M, Kucukaydin Z. Spontaneous Rupture of the Imperforate Hymen in an Adolescent Girl with Hematocolpometra. *Obstet Gynecol*. 2011;520304. doi:10.5402/2011/520304.

23. Ben Hamouda H, Ghanmi S, Soua H, Sfar MT. Spontaneous rupture of the imperforate hymen in two newborns. *Archives de Pediatrie: Organe Officiel de la Societe Francaise de Pediatrie*. 2016;23:275-278.
24. Winderl LM, Silverman RK. Prenatal diagnosis of congenital imperforate hymen. *Obstet Gynecol*. 1995;85:857-60.
25. Ogunyemi D. Prenatal sonographic diagnosis of bladder outlet obstruction caused by a ureterocele associated with hydrocolpos and imperforate hymen. *Am J Perinatol*. 2001;18:15-21.
26. Lee KH, Hong JS, Jung HJ, Jeong HK, Moon SJ, Park WH, Jeong YM *et al*. Imperforate Hymen: A Comprehensive Systematic Review. *J Clin Med*. 2019;8:56. doi:10.3390/jcm8010056
27. Lardenoije C, Aardenburg R, Mertens H. Imperforate hymen: a cause of abdominal pain in female adolescents. *BMJ Case Rep*. 2009; bcr08.2008.0722. doi:10.1136/bcr.08.2008.0722
28. Chelli D, Kehila M, Sfar E, Zouaoui B, Chelli H, Chanoufi B. Imperforate hymen: Can it be treated without damaging the hymenal structure? *Sante*. 2008;18:83–87.