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Case Report

Pyocolpos at puberty: a very rare entity

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ABSTRACT

This is a case report of a 12 year girl who reported with a H/O incomplete bladder evacuation & burning in micturition since one month, urinary incontinence once only. She had no abdominal pain or fever & had not attained menarche. The stature was short, thelarche attained 4 months back. No bulge in abdomen. Other general examination features normal. Urine examination was normal. USG suggested hematocolpos with imperforate hymen. On this basis local examination was done. Imperforate hymen with otherwise normal external genitalia found. Pubic & axillary hair absent. Preoperative diagnosis was imperforate hymen & hematocolpos with? androgen insensitivity syndrome. She could not afford serum DHEAS levels. With this diagnosis it was worrisome to think that patient had attained menarche (cryptomenorrhoea) with androgen insensitivity as pubic & axillary hair had not appeared even after the presumed menarche, which should have appeared well before menarche. Her short stature was also not expected to change after menarche. Hymenotomy under GA done, 490 ml of mucopurulent discharge was drained. Postoperative period uneventful. Now she is expected to have a growthspurt, adrenarche & menarche. In prepubertal girls with urinary complains, the possibility of imperforate hymen with hematocolpos/pyocolpos should be considered as differential diagnosis. Incorporating an examination of external genitalia into routine practice of clinicians caring for children can prevent the significant delay in the diagnosis of imperforate hymen.

Keywords: Pyocolpos, Hematocolpos

INTRODUCTION

The hymen is formed by the fusion of sinovaginal bulbs & urogenital sinus. The Mullerian ducts meet the sinovaginal bulbs at the most cephalad tip of the invaginating urogenital sinus. The canalization of vaginal plate forms vagina & canalization of most caudal portion of the vaginal plate at the urogenital sinus establishes a patent hymen. If canalization fails to occur it remains imperforate. Imperforate hymen is the most frequent cause of vaginal outflow obstruction, occurring in 0.1% of newborn girls. This can cause hematocolpos due to accumulation of menstrual blood at puberty or mucocolpos as a result of collection of cervical secretions.

Hematocolpos always occurs at puberty, but mucocolpos can occur in neonates under the effect of circulating maternal oestrogens causing cervical secretions; or at puberty, as a result of endogenous oestrogen stimulations. This can get converted to pyocolpos secondary to infection. Mucocolpos/pyocolpos is seen less commonly than hematocolpos in girls with vaginal outflow tract obstruction.

CASE REPORT

A 12 years girl was admitted at Chirayu medical college & hospital, Bhopal on 14.07.14 at 12.34 pm with presenting complains of incomplete bladder evacuation since one month & burning in micturition. There was H/o urinary incontinence once 7 days back, no history of abdominal pain or fever. She had not attained menarche.

 $\mbox{O/E}$ - weight 42 kg, short statured, height $\,$ 136 cm., no pallor, no pedal oedema, lungs clear, heart sounds

normal, abdomen soft, no mass or bulge found, breasts normally developed, tanner stage 3 (thelarche attained 4 months back). Was advised routine urine examination & pelvic USG. Urine examination was within normal limits.

Transabdominal & transperineal USG showed normal contour of urinary bladder with echofree content, uterus normal size with distended vagina & cervical lumen, endometrial echo thick with fluid seen within, intact subendometrial halo, myometrial echoes homogeneous. Large collection with particulate matter seen in vaginal & cervical lumen, measuring 90 x 60 mm? haematocolpos. Thick vaginal septum of 4.4 mm thickness present, both ovaries normal in size with immature follicles, POD free.

Ultrasonic impression - normal uterus with large hematocolpos & vaginal septum.

After the USG report examination of external genitalia was done which was not thought necessary before this.

L/E - external genitalia well developed, axillary & pubic hair not present, thick imperforate hymen at vaginal introitus, bulging outside, pink in colour.

P/R - soft anterior bulge could be palpated 3 cms above anal sphincter.

Investigations

Hb 11.6 gm%, TWBC 8700/cumm, platelets 2.10 lakh/cumm, urine routine examination - WNL, BT 2.45 min, CT 4.45 min, RBS 84 mg%, HBsAg , HIV I & II nonreactive, TSH 3.63 $\mu IU/ml$, free T_4 1.37 ng/dl. She was advised serum DHEAS & total testosterone but patient could not afford it.

Preoperative diagnosis

Imperforate hymen with hematocolpos with ? androgen insensitivity.

Procedure

Hymenotomy done under GA on 16.07.14.

Intraoperative findings

Hymen bulging, urethra normal. Hymenotomy was done. Surprisingly, there was no hematocolpos but 490 ml of mucopurulent discharge was drained. Intermittent sutures put on the edges with 4-0 vicryl. Patient stood the operation well. Collected discharge sent for C & S. No organisms grown.

Postoperative diagnosis

Imperforate hymen with pyocolpos. She was given antibiotics, perineal care & supportive treatment.

Postoperative follow-up - after two months of hymenotomy, hymen is normal with the opening patent, no discharge.



Figure 1: Imperforate hymen with absent pubic hair.



Figure 2: Developed breast and absent axillary hair.



Figure 3: Ultrasonic imaging showing distended vagina secondary to collection.



Figure 4: Pyocolpos draining.

DISCUSSION

Normal pubertal development occurs in a predictable orderly sequence over a definite time frame. When pubertal development occurs asynchronously, with development of breast in the absence of significant pubic & axillary hair, the diagnosis is usually androgen insensitivity. Although generally the first sign of puberty is accelerated growth, breast budding is usually the first recognized pubertal change, followed by the appearance of pubic hair, peak growth velocity & menarche.² In this case preoperatively the diagnosis made was imperforate hymen with hematocolpos with? androgen insensitivity. With this diagnosis it was worrisome to think that patient had attained menarche (cryptomenorrhoea) with androgen insensitivity; as pubic & axillary hair which are adrenal functions, had not appeared even after the presumed menarche, which should have appeared well before menarche. Other fact to consider was her short stature which was not expected to change after menarche.

But after the diagnosis of pyocolpos, all these worries were over. As she had thelarche four months before, the sequential appearance of adrenarche, growth spurt & onset of menarche are now well expected. On these grounds presently the adrenal function does not appear to be abnormal clinically, as menarche has not set in till

The urinary incontinence which she had, appears to be due to overflow incontinence.

This was a very rare presentation causing a diagnostic dilemma. Very few cases have been reported with pyocolpos presenting at puberty.

Indian health journal has published a report of a 7 months old girl with mucocolpos.³ Shen MC et al. have reported a mucoocolpos with lobar nephronia in a 2 year girl. If it goes undetected, it can cause complications like

hydrouretor & hydreonephrosis.^{3,4} It can present with UTL⁵

Pyocolpos has also been reported with distal vaginal atresia during infancy presenting with acute intestinal obstruction and acute urinary retention. Imperforate hymen can also occur with transverse vaginal septum. Reports are available with these two anomalies occurring in combination, causing distal mucocolpos with proximal hematocolpos or pyocolpos with abdominovaginal fistula. In the second sec

CONCLUSION

In prepubertal girls with urinary complains, the possibility of imperforate hymen with hematocolpos/pyocolpos should be considered as differential diagnosis.

Incorporating an examination of external genitalia into routine practice of clinicians caring for children can prevent the significant delay in the diagnosis of imperforate hymen.

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