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Case Report

Late diagnosis of imperforate hymen with hematometrocolpos and bilateral hydronephrosis of a horseshoe kidney☆☆☆☆

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ABSTRACT

Imperforate hymen is a fairly uncommon genital disorder where the hymen completely obstructs the vaginal opening. Several authors have ruled out its co-occurrence with other congenital anomalies. In this report, we discuss the exceptional case of a late diagnosis of imperforate hymen associated with bilateral hydronephrosis of a horseshoe kidney in a 19-year-old female patient. To our knowledge, an association of imperforate hymen and horseshoe kidney has never been reported.

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Introduction

Imperforate hymen is a female genital abnormality where the hymen completely obstructs the vaginal opening. Its occurrence is relatively rare, with an approximate incidence of 1 in 2000 female births [1]. Hematometrocolpos is a rare compli-

cation of imperforate hymen and is found in 0.14% of cases. Hematometrocolpos may lead to a mass effect on the bladder and ureter and ultimately cause urinary tract obstruction [2,3]. The association of imperforate hymen and other urogenital anomalies has been ruled out by several authors [4,5].

We report the case of a 19-year-old patient who was seen for acute abdominopelvic pain. A CT scan revealed an imper-

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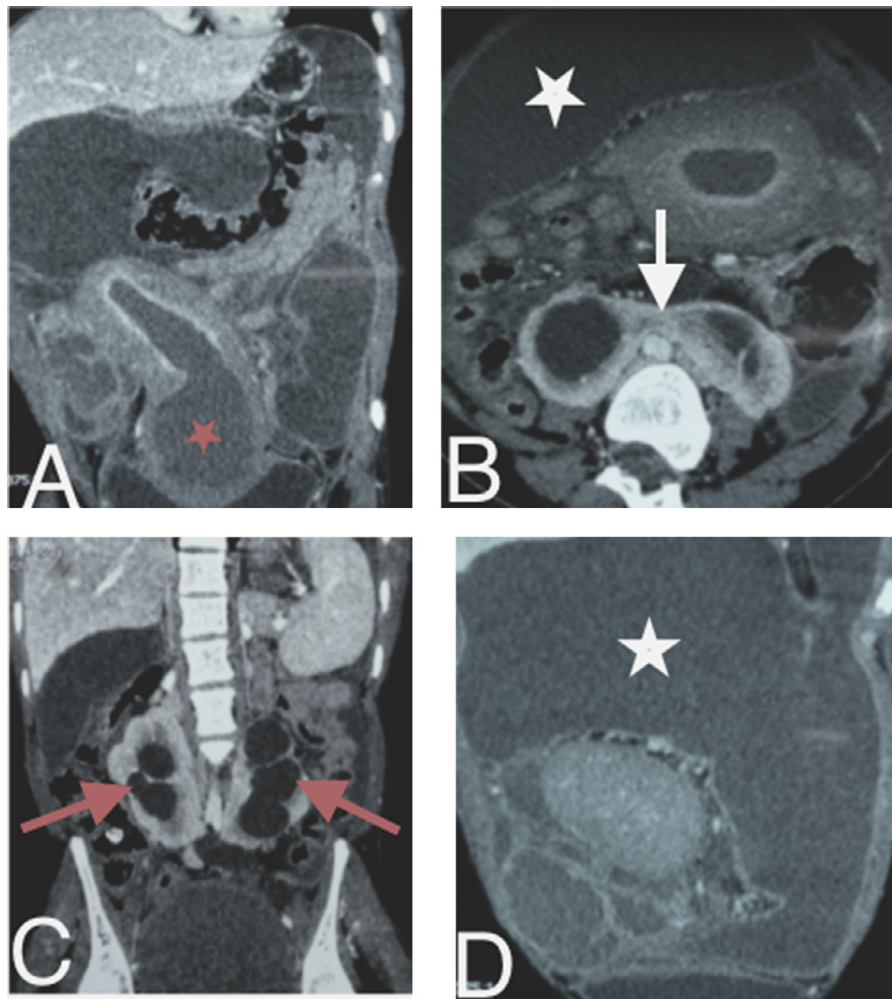


Fig. 1 – Abdominal CT scan with contrast. (A) Coronal view showing dense collection dilating uterine and vaginal cavities (red star). (B) Axial view showing the anterior loculated peritoneal collection (white star) and the horseshoe kidneys, joined by a median tissue band (white arrow). (C) Coronal view showing renal cavity dilation (red arrows) causing bilateral hydronephrosis. (D) Coronal view showing the dense peritoneal collection (white star).

forate hymen associated with horseshoe kidneys, complicated by a hydronephrosis and a hematometrocolpos.

To our knowledge, an association of imperforate hymen and horseshoe kidneys has not been reported in the literature.

Case report

This is a 19-year-old female patient, nulliparous and nuligravida, with a long history of evolving and acute abdominopelvic pain. The patient also reported episodic paroxysmal pain.

The patient had normal vital signs, and a low-grade fever with a body temperature of 38°C. A clinical examination showed a severe abdominal distension and an abdominopelvic mass.

An abdominal CT scan with contrast revealed an important uterovaginal distension that resulted from the collection of a dense fluid (Fig. 1A); this is consistent with a hematometrocolpos caused by imperforate hymen.

The hematometrocolpos caused a mass effect on the posterior wall of the bladder which led to a bilateral ureterohydronephrosis (Fig. 1C) in a horseshoe kidney (Fig. 1B). In addition, a loculated fluid collection in the peritoneal cavity was noted (Figs. 1B and D).

With further exploration, we learned of the patient's history of amenorrhea and multiple consultations for cyclic pelvic pain, and a lack of improvement after medical treatment. A gynecological examination showed a membrane completely obstructing the vaginal opening, thus confirming the diagnosis of imperforate hymen.

An exploratory laparotomy showed a uterovaginal distension, associated with a bilateral hydrosalpinx. In addition, there was a thick, encapsulated intraperitoneal collection, covered with peritoneum. The collection had a dark brown color.

An aspiration with peritoneal toilet and adhesiolysis, and an incision of the posterior wall of the vagina were performed. A liquid similar to that of the peritoneal fluid was found. A vaginal toilet was completed, followed by a vaginoplasty.

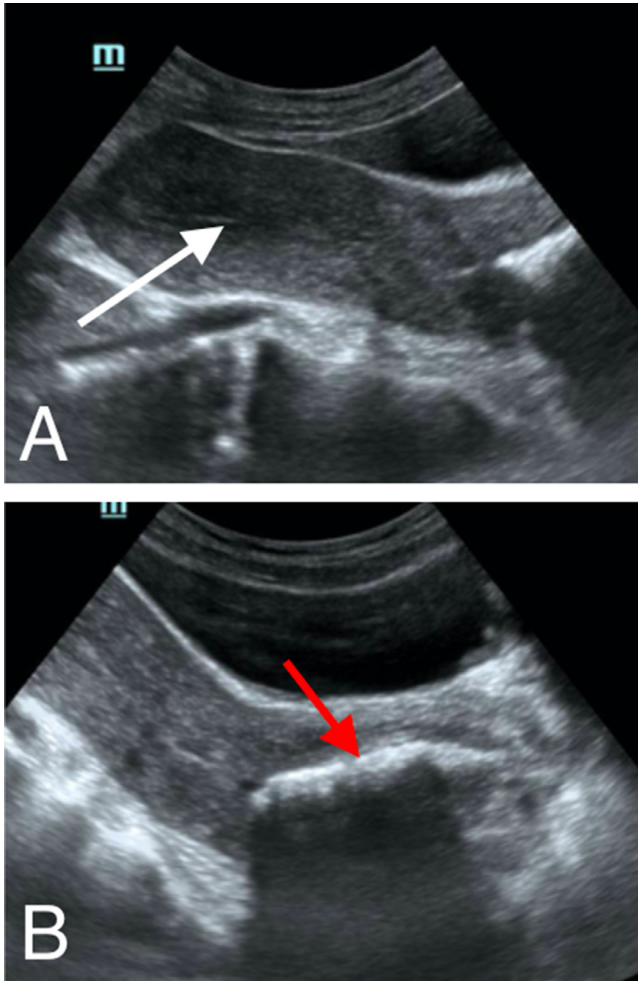


Fig. 2 – Transabdominal pelvic ultrasound. (A) longitudinal section passing through the uterus, showing complete evacuation of the uterine cavity (white arrow). (B) longitudinal section passing through the vagina showing an empty cavity of fluid but containing air bubbles responsible for a posterior artifact (red arrow).

The postoperative course was uneventful. At the 5-month follow-up, the patient reported that she had normal menstrual periods, without dysmenorrhea; each cycle lasted for 4 days. A transabdominal ultrasound control was performed, and it showed a complete evacuation of peritoneal, uterine and vaginal cavities (Fig. 2).

Discussion

Imperforate hymen is a rare disease with an incidence of 1 in 2000 female births [1]. It is a congenital condition for which antenatal diagnosis is possible but rare [6]. Most often, it is diagnosed in adolescence after an occurrence of cryptomenorrhea which presents as cyclic pelvic pain associated with amenorrhea.

For our patient, the diagnosis was delayed since it happened in her adulthood, and that despite a strong clinical his-

tory and multiple consultations. This delay in diagnosis could be explained by consultations that she had with nonspecialists, but also because clinicians do not always perform gynecological physical examination, especially if the patient is young and/or virgin.

To overcome late diagnosis, gynecological physical examination should be systematic in cases of abdominal pain, regardless of the age or virginity of the patient, especially when there is suggestive clinical history of cyclic pain.

Urinary tract obstruction is a common complication of imperforate hymen, found in 20%-48% [3,7] of cases. This urinary obstruction may, in rare occasions, lead to bilateral hydronephrosis brought about by the mass effect of hematometocolpos of the ureter [3].

The incidence of hydronephrosis in horseshoe kidneys is higher than in normal kidneys. This can be explained by the anomalous structure of a horseshoe kidney, and how that anomaly is conducive to the obstruction of the urinary tract [8].

Without adequate treatment, hydronephrosis can result in kidney failure.

For our patient, renal function was preserved despite a bilateral hydronephrosis.

The reported associations of imperforate hymen with other congenital anomalies are not relevant to urogenital anomalies. In fact, for some authors, it is not necessary to systematically look for it [4,5]. For our patient, the association of imperforate hymen and horseshoe kidney may have been coincidental, and this has been the first reported case to our knowledge.

The presence of a dark brown peritoneal effusion similar to an endovaginal collection could have been caused by either the perforation of the hematometrosalpinx or by retrograde bleeding [9].

In our patient's case, since a surgical exploration showed that her fallopian tubes were dilated but intact, retrograde bleeding was more probable. In the long run, this could lead to endometriosis and hypo fertility [10]. Our patient would benefit from fertility consultations in the future.

Author contribution

IN wrote the manuscript with input from and KND et AN.

IN, MT, and CTD edited the illustrations.

SK and KND are following up with the patient.

SB and SK supervised the work.

All the authors have read and approved the final version of the manuscript.

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