Does Menstrual Flow Exclude Hematometra? A Rare Case of Uterine Anomaly Presenting With Anorectal Malformation

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Hematometra, which is defined as accumulation of menstrual secretions in the uterine cavity, may not be diagnosed until the maturating adolescent fails to menstruate. Clinically, a lower abdominal mass and periodic abdominal pain may develop in these children after puberty. Here, a 13-year-old girl with menstrual flow who presented with symptoms of genital outflow tract obstruction is described.

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FAILURE of the female genital tract to develop may result in genital outflow tract obstructions. The most common causes include imperforate hymen, transverse vaginal septum, vaginal agenesis, and cervical agenesis. Because there is no outlet for menstrual flow, endometrial secretions accumulate in the vagen (hematocolpos) and uterus (hematometra), respectively, with the onset of menstruation.[1] A rare case of hematometra in a 13-year-old girl is described after surgical correction of anorectal malformation.

CASE REPORT

We report a case of a 13-year-old girl who had presented with a history of lower abdominal pain, vomiting, and fever on arrival at a state hospital. The pain had been vague and continuous for a week, accompanied by bilious vomiting. Her menses had started a few months before, which was considered normal in duration and amount, except for significant dysmenorhaea.

Medical reports also state that at birth she had intermediate anorectal malformation, including anal stenosis, a simply narrowing anus at its normal location with normal sphincteric function, and rectovaginal fistula. The opening of the rectovaginal fistula was observed on the upper posterior vaginal wall. The child was able to pass stool both from the vagina and anal canal, and did not require any dilatations to assist with defecation. A left sigmoid diverting colostomy, fistula division, and anoplasty was performed at the age of 6 months. Neither laparotomy nor radiography showed any evidence of uterine anomaly on medical reports. The colostomy was taken down a year later. The child has not had any defecation problems since repair.

Reportedly, physical examination revealed a slightly mobile lower abdominal mass and tenderness. Roentgenographic studies showed displacement of the bowel by the pelvic mass. Ultrasonography indicated a regular, 12- × 10- × 10-cm cystic mass that was pushing the rectum and the uterus to the left. Computed tomography scanning confirmed these findings. The case was interpreted as a complicated pelvic-abdominal mass, necessitating explorative laparotomy. Laparotomy is reported to have shown intensely indurated pelvic tissues and a cystic mass, adjacent to the uterus, containing dark brown material. The anatomic limits were difficult to define, and to prevent pelvic injury, just viscous fluid was suctioned, and a drain was left in the space. She was treated postoperatively by intravenous fluids, nasogastric decompression and antibiotics. However, the drainage was still present a month after the operation when she was referred to our hospital.

On admission, her acute complaints had been resolved. The drain was still present on the lower right quadrant. The mass had diminished and was not palpable on physical examination. Rectal examination showed a firm mass anterior to the rectum. Radiographic imaging showed similar findings (Fig 1) to the medical records of the referring center. After preoperative evaluation, reexploration was undertaken. Laparotomic examination revealed resolved pelvic peritoneal inflamation. However, it showed significant, dense fibrous adhesions. The mass could not be appreciated as separate and distinct from the right ovary. The residual cavity of the mass had no outlet to any pelvic tissue and was removed. Histopathologic examination showed proliferative endometrium and an ovarian endometriotic cyst. The patient was discharged free of complaints on the seventh day after the operation. Currently, her menstrual periods are normal, and the preoperative symptoms are completely resolved.

DISCUSSION

Various congenital anomalies can be found in association with anorectal malformations.[2] In rare instances, girls are found to have duplication of the genitalia.[3] The arrested descent of the urorectal septum, which normally separates the genitourinary tract from the gastrointestinal tract, interferes with fusion of the Müllerian ducts. Failure of fusion of the Müllerian ducts may result in double a uterus with separate vagina. This clinical entitiy is asymptomatic because there is an outlet for menstrual secretions. However, rarely, asymmetric bicornuate uterus with a rudimentary horn that does not communicate with the vagina

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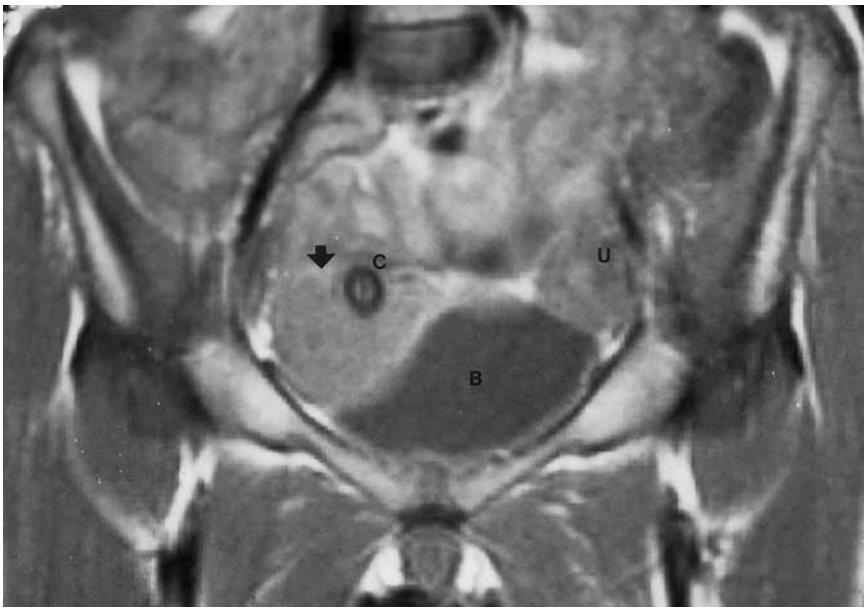

Fig 1. T_1-weighted coronal magnetic resonance image of the pelvis indicates a right abdominal mass (arrow) with an indwelling catheter (C) causing indentation in the bladder (B). Another soft tissue intensity is seen on the left side of the pelvis whose configuration and signal characteristics are in accordance with the uterus (U).

will become symptomatic with the onset of menstruation.[4] Obstructed uterovaginal anomalies typically manifest after puberty. The most common symptoms are lower abdominal pain radiating to the lower back, discomfort, and sense of fullness in the pelvis. A suprapubic tender mass often is palpable as a result of uterine enlargement and upward displacement. Urinary retention or constipation can occur because of compression of the distended uterus.[5]

When a girl has a pelvic mass with equivocal physical findings, the question of obstructed uterovaginal anomaly always is raised, especially if she has periodic severe dysmenorrhea after puberty with no onset of menarche. Despite monthly cycles, this girl presented with symptoms of genital outflow tract obstruction. Because she had unilateral obstruction, there was cyclic bleeding from the hemiuterus and vagina of the unobstructed side and obstructive symptoms with development of hematometra from the other side.[6] The increasing pressure of the obstructed side may have caused retrograde passage of blood freely into the peritoneal cavity via the salpenx-forming hematoperitoneum. This can explain the sign and symptoms of peritoneal irritation necessitating urgent explorative laparotomy. There was evidence of thelarche, adrenarche, and ovarian estrogen production.

Neither at sonographic evaluations nor at previous operations had the obstructed half of the genital tract been identified. This can be related to the small outer circumference of the rudimentary horn before puberty. However, the onset of endometrial secretions resulted in uterine enlargement presenting the acute clinical syndrome.

Because of the severity of symptoms and long-term complications, such as infertility, infection, and endometriosis, genital outflow tract obstruction remains one of the few conditions for which treatment should not be delayed once the diagnosis is made in an adolescent female.[6] Excision is the only therapy for a rudimentary obstructed uterine horn.[4] Detailed history and a careful physical examination, even in emergency situations, are essential. Hematometra always should be a differential consideration when a cystic midline mass is identified arising from the pelvis.[1] Clinical suspicion with recognition of the clinical syndrome, particularly in anorectal malformations, is essential in diagnosing this disease.

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