

Transverse vaginal septum: a benign reason for elevated serum CA 19-9 and CA 125 levels

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Introduction

Transverse vaginal septum is relatively rare anomaly, occurring in about 1 in 70,000 girls [1]. It is a developmental defect in the embryogenesis of the vagina that leads to incomplete fusions between the müllerian duct component and the urogenital sinus component of the vagina [2]. The patients complain about hydrocolpos, primary amenorrhea and cyclic pelvic pain due to the development of hematocolpos [3].

Tumor markers such as CA19-9 and CA125 are commonly used markers in the diagnosis of pelvic masses. CA19-9, a side branch of the Lewis blood group system, is a sialylated Lewis A antigen that is highly expressed by many adenocarcinomas of the digestive tract. The CA125 is expressed in coelomic epithelia such as müllerian epithelium, peritoneum, pleura and pericardium [4].

In the literature, transverse vaginal septum is not considered as a benign gynecological disorder that causes elevation of CA19-9 and CA125. By presenting this case, we wanted to mention about vaginal anomalies that cause elevation of tumor markers with a literature review.

Case report

A 15-year-old girl was admitted to our gynecology clinic with a history of cyclic pelvic pain in the lower abdomen during the last 3 months and the pain had increased in the

previous 2 days. Although secondary sexual characteristics that had already developed and were normal, menarche had not started yet. An ultrasound revealed a mass with a measure of 79×61 mm in the vagina compatible with a large hematocolpos, which continued to the cervix and normal ovaries and kidneys. Pelvic examination revealed normal perineum and intact annular hymen. A tender soft mass was palpated in the upper part of the vagina on rectal examination. Magnetic resonance imaging examination revealed a 9×7 cm cystic-like structure in the pelvis compatible with a severely dilated vagina and a hyperintense image representing transverse vaginal septum with thickness of 3 cm (Fig. 1). Total vaginal length was 2 cm measured using a hystrometer and vaginal septum was detected on the distal third of the vagina. No abdominal masses were palpated. The remainder of the physical examination was normal. The tumor markers were elevated which was CA19-9: 40.9 ml and CA125: 80.2 U/ml. Serum FSH, LH, estradiol, and prolactin levels of the patient were within normal range. Additional medical and surgical histories were unremarkable. The patient and her family were informed about surgical and expectant management and they refused any vaginal or abdominal surgery because of cultural values that give importance for the hymenal integrity representing virginity.

Discussion

Embryologically, the sinovaginal bulbs invaginate from the urogenital sinus and meet the müllerian tubercle on the caudal end of the müllerian ducts to form the vaginal plate that is then canalized to form the lower part of the vagina. Failure of the vaginal canalization results in the transverse vaginal septum [5]. This septum may lie at the level of

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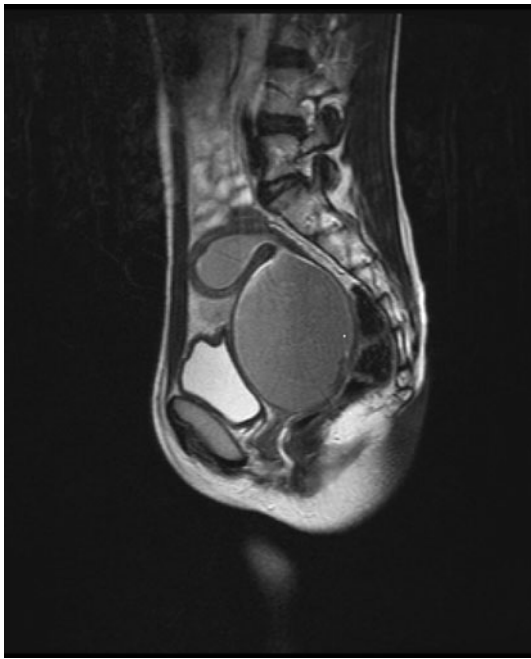


Fig. 1 MRI findings in a patient with transverse vaginal septum

upper, middle or lower third of the vagina in the frequencies of 46, 35 and 19 %, respectively, with the septum getting thinner as it gets closer to the introitus [6]. The clinical appearance of patients with transverse vaginal septum varies including a double ureter, a rib malformation, a unicornuate uterus, and/or imperforate hymen [7]. Ultrasonography and magnetic resonance imaging are helpful to delineate the septal position and thickness so as to plan the surgery appropriately [6].

Several surgical treatment modalities have been developed to treat the transverse vaginal septum and it varies from simple incision to surgical excision followed by approximation with the pull-through or the push-through technique of the corresponding portions of the transversely cut edges of the upper and lower mucosal membranes of the septum [2, 8]. To decrease the risk of post-operative stenosis, a Z-plasty alone or Z-plasty with Grünberger modification may be performed [2]. Beyth et al. [9] presented an alternative management of the transverse vaginal septum surgery, after 5 years of expectant management. Postoperative conditions, such as vaginal stenosis and re-obstruction, are major restrictive complications especially when the septum is thick [1]. In our case, the family and the patient had informed about treatment options and possible complications. After all they refused any surgical interventions and their major concern was about the defloration of the hymen that representing virginity according to their cultural values.

CA19-9 is a tumor marker for pancreatic, gastric, colonic and mucinous ovarian carcinoma [10]. However, several benign conditions such as pancreatitis, pancreatic

cysts, pancreatic or cholangial duct stenosis due to gallstones, cholangitis, papillitis, chronic hepatitis, chronic glomerulonephritis, diabetes mellitus, hemodialysis or peritoneal dialysis, bronchitis, bronchial, cysts, bronchiectasis, pulmonary fibrosis, ovarian cysts, endometriosis and finally pregnancy can be associated with elevated serum CA19-9 levels [11]. CA 125 is an antigenic determinant and is normally expressed in mesothelial cells of pleura, pericardium and peritoneum and in tubal, endometrial and endocervical epithelium [10]. It is used for screening of ovarian, pancreatic, breast, colon and lung cancers. It also increases in some benign and physiological conditions like pregnancy, fibroids, menstruation and endometriosis [11]. In our case, the tumor markers were studied for differential diagnosis of benign or malign adnexal masses. Table 1 shows the tumor markers in genital malformations.

In a previous study by Deligeoroglou et al. [7], four cases of transverse vaginal septum were reported. In these cases they had evaluated tumor markers in two patients. One of these patients had elevated only CA125 level which was 48 U/ml and the other one had elevated both CA19-9 and CA125 levels which were 1,552 and 195 μ /ml, respectively. In our case, we had also chance of the results of both elevated CA19-9 and CA125 levels being 40.9 ml and 80.2 U/ml, respectively. Partsinevelos et al. [10] and Buyukbayrak et al. [11] also reported elevated CA19-9 and CA125 levels in cases with imperforate hymen. Kalmantis et al. [12] reported only elevated CA125 level in a case of imperforated hymen.

According to these findings in case of obstructive genital disorders, such as transverse vaginal septum and imperforate hymen, we may postulate that elevated CA19-9 levels may be due to the proliferation of non-cancerous tissue or obstruction of CA19-9 discharge pathways and elevated CA125 levels may be due to increased release from endometrial epithelium.

Table 1 Tumor markers in genital malformations

Study	Age	Diagnosis	CA19-9	CA125
Deligeoroglou et al.	13	Transverse vaginal septum	1,552 μ /ml	195 μ /ml
Deligeoroglou et al.	12	Transverse vaginal septum	–	48 μ /ml
Buyukbayrak et al.	13	Imperforate hymen	>1,000 U/ml	457 U/ml
^a Partsinevelos et al.	12	Imperforate hymen	960 U/ml	277 U/ml
Kalmantis et al.	15	Imperforate hymen	–	70 U/ml
Present study.	15	Transverse vaginal septum	40.9 ml	80.2 U/ml

^a The tumor markers were evaluated postoperative third day

In conclusion, the presence of a transverse vaginal septum should be considered in cases of abdominal pain, primary amenorrhea, and hematocolpos in adolescent girls. Informed consent has to be obtained from the family and the patient about surgical procedures. Although the information about elevated tumor markers in obstructive genital disorders is limited with the case reports, further studies with large series has to be performed about this issue.

Conflict of interest We declare that we have no conflict of interest.

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