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CASE REPORT

Imperforate hymen: a new benign reason for highly elevated serum CA 19.9 and CA 125 levels

Esra Esim Buyukbayrak · Ayse Gul Ozyapı · Yasemin Karageyim Karsıdag · Zehra Meltem Pirimoglu · Orhan Unal · Cem Turan

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Absract Imperforate hymen is a urogenital tract anomaly that represents the most frequent congenital malformation of the female genital tract. CA 19-9 and CA 125 are widely used as tumor markers, however several benign conditions are also known to increase serum CA 19-9 and CA 125 levels. According to classical textbook knowledge, imperforate hymen is not listed under the benign conditions that increase serum CA 19-9 and CA 125 levels. In this case report we describe a relation between imperforate hymen and elevated serum CA 19-9 and CA 125 levels.

Keywords Imperforate hymen · CA 19-9 · CA 125

Introduction

The hymen is a mucous layer of epithelized connective tissue that separates the lumen of the vagina from the cavity of the urogenital sinus during fetal life. This membrane usually ruptures and is partially reabsorbed during the later stages of embryo development. Failure of this process results in a congenital anomally called "imperforate hymen".

Imperforate hymen is a urogenital tract anomaly that represents the most frequent congenital malformation of the female genital tract. It is a sporadic disease with an incidence that ranges between 0.014 and 0.1% at term [1].

E. E. Buyukbayrak · A. G. Ozyapı · Y. K. Karsıdag · Z. M. Pirimoglu · O. Unal · C. Turan
Dr. Lutfi Kırdar Kartal Education and Research Hospital, Istanbul, Turkey

E. E. Buyukbayrak (✉)
Dr. Zeki Zeren sok, No: 6/24, Goztepe, Istanbul, Turkey
e-mail: esraesim@yahoo.com

Generally, imperforate hymen does not show symptoms until puberty, when cervical and vaginal secretions and menstrual blood lead to distention of the vagina (hydrocolpos) and of the uterine cavity (hydrometracolpos).

CA 19-9 is widely used as a tumor marker for cancers of the pancreas, stomach, colon, cholangial duct, ovaries, endometrium, and lung (adenocarcinoma). However, several benign conditions are also known to increase serum CA 19-9 levels [2].

CA 125 is an antigenic determinant widely used for screening of ovarian, pancreatic, breast, colon and lung cancers. It also increases in some benign and physiological conditions like pregnancy, menstruation and endometriosis [2].

In this case report we present an imperforate hymen case that is accidentally evaluated first for abdominal mass with highly elevated tumor markers, which is then followed up for tumor marker levels.

Case

A 13-year-old premenarchal girl who has urinary retention, abdominal distention and discomfort complaints has admitted to general surgery department on 30 May 2006. Her clinical examination revealed a palpable abdominal mass and general surgery department asks for tumor markers and abdominal MRI. Her tumor markers were highly elevated (CA 19-9 > 1,000 U/ml, CA-125: 457 U/ml, CA 15-3: 31 U/ml, CEA: 1.30 ngr/ml) and her abdominal MRI revealed all intraabdominal organs normal except for 15 × 8 × 10 cm. Hydrometrocolpos appearance without accompanying uterine anomaly (Fig. 1). Then the patient was consulted to our gynecology clinic on 6 June 2006. Her genital examination confirmed imperforate grayish purple hymenal membrane at inspection and hydrometrocolpos

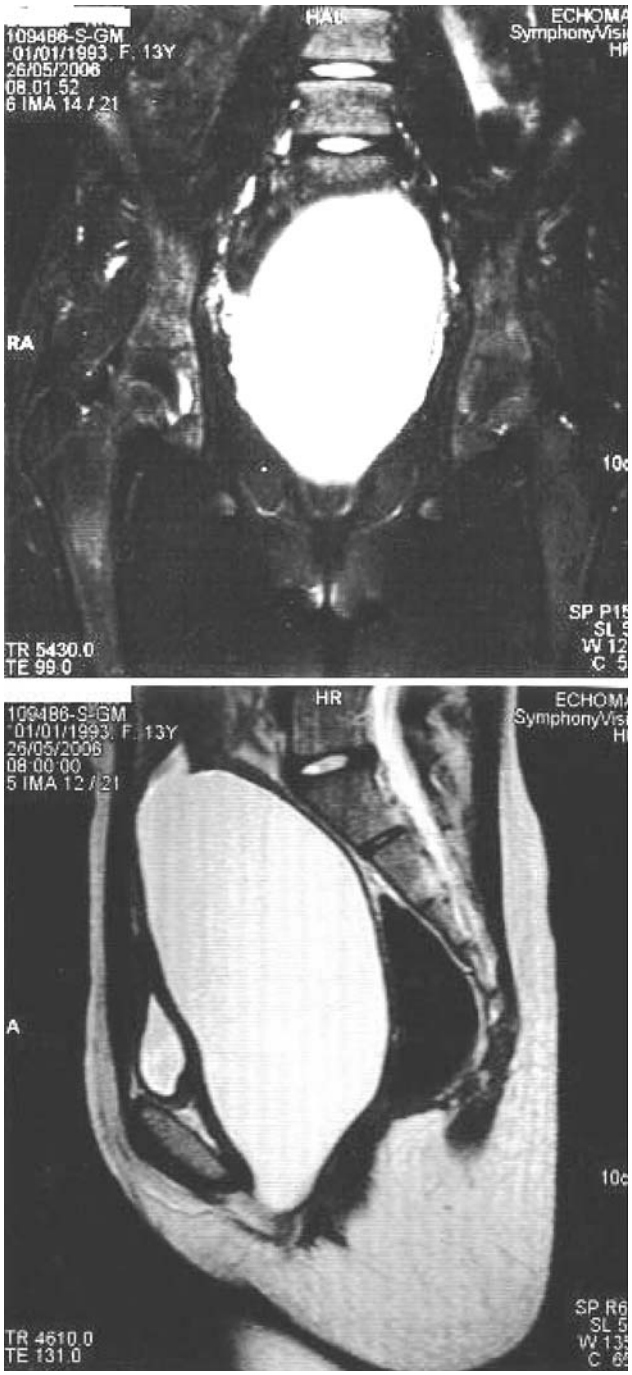

Fig. 1 Coronal and sagittal section of MRI view of hydrometrocolpos

appearance on ultrasonographic examination. Next day under general anesthesia we performed surgical treatment with T incision of the hymen followed by immediate leakage of 300 ml of vaginal and uterine hemorrhagic collection. After surgery she had no complaints. After hospital discharge we keep follow up for the elevated tumor markers. After one week from the operation her CA 19-9 level was 270 U/ml, CA 125 level was 121 U/ml and CA 15-3 level was 23 U/ml. After two weeks from the operation her CA 19-9 level was 19 U/ml, CA 125 levels was 17 U/ml and CA15-3 level was 16 U/ml (Fig. 2).

Discussion

In the embryological period, the lateral portion of the hymen originates from a fold of urogenital sinus, at the union of Müller's duct, while in its posterior part it originates from the cells of the urogenital sinus, externally, and from Müller's duct, internally. Usually in the 8th week of gestation, it partially ruptures in the inferior part of Müller's duct, remaining as a fold of mucous membrane around the entrance of the vagina. Failure of these events results in a persistence of the septum (imperforate hymen). Because of this vaginal outflow obstruction, there may be significant accumulation of cervical and vaginal secretions, resulting in hydrocolpos and hydrometrocolpos [3].

CA 19-9 is widely used as a tumor marker for cancers of the pancreas, stomach, colon, cholangial duct, ovaries, endometrium, and lung (adenocarcinoma). However, several benign conditions are also known to increase serum CA 19-9 levels.

In one group of benign conditions, inflammation or proliferation of noncancerous tissue results in elevated serum CA 19-9 levels. Pancreatitis, pancreatic cysts, cholangitis, bronchial cysts, bronchiectasis, pulmonary fibrosis, ovarian cysts, endometriosis, and pregnancy are known to be such conditions. These conditions sometimes result in a very high level of CA 19-9 (greater than 1,000 U/ml).

In a second group of benign conditions, serum CA 19-9 levels are elevated by an obstruction of CA 19-9 discharge

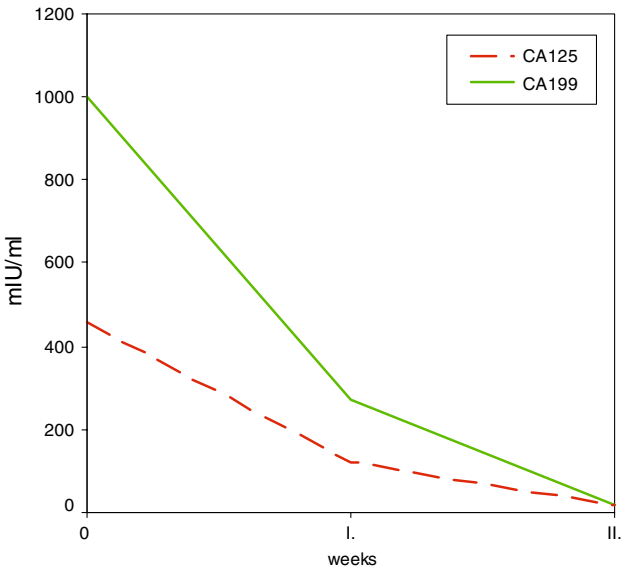

Fig. 2 CA 19-9 and CA 125 levels according to time (weeks after the operation)

pathways. Pancreatic or cholangial duct stenosis due to gallstones, papillitis, and bronchitis are such conditions.

In the third group of benign conditions, such as chronic hepatitis, chronic glomerulonephritis, diabetes mellitus, and possibly hemodialysis or peritoneal dialysis, malfunction in organs that metabolize CA 19-9 trigger an elevation in serum levels of CA 19-9 [4].

CA 125 is expressed by amniotic and coelomic epithelium during fetal development. In adult, it is found in structures derived from coelomic epithelium (the mesothelial cells of pleura, pericardium and peritoneum) and in tubal, endometrial and endocervical epithelium [2].

When we look at the literature related with imperforate hymen most of the publications are about clinical presentations, urinary retention, and treatment strategies. Our case is the first case in the literature showing that imperforate hymen can be one of the benign conditions that can increase CA 19-9 and CA 125 levels.

The mechanism that leads to highly elevated CA 19-9 level in imperforate hymen condition can be either inflammation of the tissue or obstruction of discharge pathway.

Elevated CA 125 level in imperforate hymen condition can be explained by expression of CA 125 in endocervical epithelium and desquamation of this epithelium in imperforate hymen content.

To be able to list imperforate hymen under the benign conditions that cause elevated serum CA 19-9 and CA 125 levels more cases are needed but we think that there is a strong relation between them.

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