

Uncommon Elevation of CA 19-9 in Dermoid Cyst of the Ovary: A Case Report

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ABSTRACT

Cancer antigen 19-9 (CA 19-9), a tumor marker, has been reported to be elevated in patients with pancreatic, gastric and hepatobiliary malignancies. We present an unusual case of a 21-year-old unmarried girl with dermoid cyst of the ovary with abnormally elevated CA 19-9 levels. Elevated serum CA19-9 is not an uncommon finding in patients with dermoid cyst of the ovary and could be used as a marker in the differential diagnosis in patients with suspected pelvic malignant mass.

Keywords: Malignancy, Mature cystic teratoma, Tumor marker.

INTRODUCTION

Tumor markers are substances, often proteins, that are produced by the cancer tissue itself or sometimes by the body in response to cancer growth. These markers help to detect and diagnose some types of cancer, predict and monitor a patient's response to certain treatments, and detect recurrence. CA19-9 is a sensitive marker for pancreatic, gastric and hepatobiliary malignancies. Markedly raised levels are almost always associated with advanced stage of malignancy.^{1,2} Elevated CA 19-9 has also been associated with dermoid cyst (mature cystic teratoma) of the ovary on a few occasions.³ We present herein an unusual case of a 21-year-old unmarried girl with dermoid cyst of the ovary with abnormally elevated CA 19-9.

CASE DESCRIPTION

A 21-year-old unmarried girl presented in the out-patient department (OPD) with pain and heaviness in the lower abdomen of two months' duration and scanty menses for one-year. Per abdominal examination revealed a large, mobile, non-tender, regular, 15 cm × 12 cm mass in the lower abdomen, with no evidence of ascites or organomegaly. The uterus could be palpated separately on per rectal examination. Ultrasound showed the appearance of a diffusely echogenic mass with a mural nodule which was suggestive of mature

cystic teratoma. Magnetic resonance imaging (MRI) of the abdomen confirmed the diagnosis. However, considering the age of the patient and the large size of the mass, a differential diagnosis of a malignant neoplasm was considered. Tumor markers specific to malignant ovarian neoplasms including germ cell tumors were tested. Cancer antigen 19-9 (CA 19-9) was detected to be markedly raised to 9265 U/mL (normal level 0-37 U/mL) and carcinoembryonic antigen (CEA) levels were borderline elevated 6 ng/mL (normal level 0-5 ng/mL). Other tumor markers tested including cancer antigen 125 (CA 125), lactate dehydrogenase (LDH) and alpha fetoprotein (AFP), were within normal range. A staging laparotomy revealed 18 cm x 10 cm left ovarian mass which was predominantly cystic and well encapsulated. Uterus, bilateral fallopian tubes, right ovary and rest of the pelvic and abdominal structures were normal and there were no enlarged retroperitoneal lymph nodes. A left ovarian cystectomy was performed. Cut-section revealed mature elements like hair and cartilage suggestive of a dermoid cyst and the frozen section as well as final histopathology report confirmed the diagnosis of a mature cystic teratoma. The patient made an uneventful recovery and the CA 19-9 and CEA levels returned to normal levels of 7.04 U/mL and 1.23 ng/mL respectively, on postoperative day 13. The patient was followed up in the OPD monthly over the next six months and she remained asymptomatic.

DISCUSSION

Koprowski, *et al.* first described CA 19-9 antigen in 1981.¹ It is a monosialoganglioside secreted by mucinous tumors of the gastrointestinal tract, predominantly those of the pancreas and biliary tract. It has been found to be raised in many malignancies, including colorectal carcinoma, pancreatic adenocarcinoma and epithelial ovarian carcinoma. Studies have found that markedly high levels of CA 19-9 antigen (greater than 1000 U/mL) are almost always associated with malignant tumors.² However, uncommonly such high CA 19-9 levels have been reported in certain benign conditions including acute cholangitis and cirrhosis.²

Dermoid cyst or mature cystic teratoma (MCT) of the ovary is the most common germ cell tumor of the ovary in women of reproductive age group. MCTs constitute 10-25% of all ovarian neoplasms and 60% of all benign ovarian tumors. The heterogeneous histological composition of these tumors is probably responsible for the occasional elevation of tumor markers such as CA 19-9 and CA 125. In MCTs, tumor markers CA19-9 and CA125 have been reported to be elevated in 39 to 59% and 13.5 to 25% of cases, respectively.³ Published literature has reported the association between elevated CA 19-9 level and MCT on a few occasions. In a study consisting of 215 cases, CA 19-9 levels were elevated in 40% of selected population with mature cystic teratoma; the mean serum levels of CA 19-9 were 83.5 (± 179.2) U/mL.⁴ In another study including 250 patients, 31 cases had CA 19-9 above 101 U/mL.⁵

Some studies have reported alarmingly high levels of CA19-9, raising a high index of suspicion for a malignancy, despite imaging studies and final histopathology reports being suggestive of MCT. Pyeon, *et al.* reported CA 19-9 levels of 2753 U/mL in a 37-year-old parous Korean woman.⁶ Sampaio, *et al.* reported CA 19-9 levels of 8922.75 U/mL in a 50-year-old perimenopausal woman with MCT who was treated on the lines of ovarian malignancy.⁷ Our case was unique in that unusually high levels of CA 19-9 and CEA were seen in a very young unmarried girl. In one such case of a 14-year-old from Iran, salpingo-oophorectomy had to be performed because of ovarian cyst torsion and necrosis.⁸

Few studies have reviewed the association between elevated serum CA19-9 and specific clinical features of MCTs. Dede, *et al.* retrospectively evaluated 80 cases of MCTs and concluded that patients with elevated CA19-9 levels have greater tumor size and significant relationship with bilaterality.⁹ Cho, *et al.* reviewed 239 patients with pathologically confirmed MCTs, and demonstrated a significant correlation between CA19-9 level, tumor size and fat component.³ The high serum CA 19-9 levels have been attributed to the leakage of CA 19-9 from the cyst wall into the bloodstream, as a consequence of rupture of the cyst due to

its weakened wall caused by the large diameter of the lesion.^{8,10} The elevated levels may also be related to ovarian torsion and to the extent of the necrosis of the ovary.⁸ Published studies have also demonstrated that an isolated elevation in the levels of CA 19-9 in MCTs is not related to malignant transformation.⁷ CA 19-9 levels can also be used for postoperative follow-up and as a marker for recurrence of mature cystic teratomas.

We conclude that abnormally high levels of CA19-9 may be associated with ovarian neoplasms. In cases of ovarian dermoid, this may be attributed to the large size of the mass, leakage from weakened cyst wall and sometimes torsion. The clinical features, imaging studies and antigen testing of dermoid cysts should be scrupulously interpreted and should not preclude a minimally invasive surgical approach as it reduces the morbidity to the patient.

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