Non-Specific increase in Ca 19.9 Level in A 17-Year-Old Patient With An Ovarian Dermoid Cyst –Case Report with Review of Literature

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ABSTRACT

Mature cystic teratomas, including dermoid cysts, are among the most commonly found benign tumors of the ovary. Malignant transformation of mature cysts occurs very rarely, usually in older patients. In this article, we presented a case report of a 17-year-old girl with a mature teratoma and a non-specifically increased level of the Ca 19.9 marker. We also reviewed the current literature. Based on the gathered data and having analysed our own case, we concluded that an increase in Ca 19.9 level is independent of the size of the tumor and its bilaterality. Moreover, it is not associated with the presence of malignant tumors. To date, the highest observed concentration of Ca 19.9 in a benign dermoid cyst has been 25590 U/mL. We concluded, that an isolated increase in the level of the non-specific Ca 19.9 marker in patients with dermoid cysts is not associated with a malignant transformation. Therefore, we recommend that in young patients the excision of dermoid cysts should be performed laparoscopically regardless of the level of the marker.

KEYWORDS: Adolescent, Cystisdermoidalis, Laparoscopy, Marker Ca 19.9, Prolactinoma.

INTRODUCTION

Dermoid cysts account for 60% of all benign ovarian tumors. In 10% of cases, they occur bilaterally, and if unilateral, they are found more frequently in the right adnexa (up to 70%). One to 3% of dermoid cysts undergo a malignant transformation, especially in older patients [1, 2]. In 15% of cases, a dermoid cyst may be complicated by ovarian torsion, and rarely, probably because of a thick cystic wall, it may rupture [3].

The presence of dermoid cysts may result in increased levels of certain markers commonly regarded as tumor markers. Most frequently (38.8%) the Ca 19.9 marker is raised, followed by Ca125 (25% of cases) [4]. The Ca 19.9 antigen, a glycoprotein, is a modified (sialated) Lewis blood group antigen. One should also remember about other causes of an increased level of Ca 19.9 - liver cirrhosis, pancreatitis, cystic fibrosis and pneumonia.

CASE REPORT

A 17-year-old girl was admitted with oligomenorrhea to the Clinic of Pediatric and Adolescent Gynecology. An ultrasound scan revealed: normal size of the body of the uterus; endometrial thickness of 5.9 mm; a non-vascularized, mixed solid-fluid lesion (80.3 x 68.9 mm) on the right; the left ovary dimensions – 30.8 x 22.4 mm, with the maximal follicular diameter of 8.6 mm. In laboratory results, a marked increase in the level of the Ca 19.9 marker was noted (up to 2333.36 U/mL), with other parameters found within their respective normal ranges (Ca125 – 29.8 U/mL, CEA – 1.12 ng/ml, AFP – 2.11 IU/ml, betaHCG < 0.1 mIU/mL, HE4 – 46.9). A significantly increased level of PRLI was also noted (up to 198.3). The value of PRL II was 307.
It was decided to urgently perform further imaging and endocrine diagnostic tests. Based on the physical examination, MRI scan and additional test results, a prolactinoma was diagnosed. Standard treatment with dopaminergic agents was initiated. Because of the young age of the patient, laparoscopic surgery was performed despite the markedly increased Ca 19.9 level. The surgery revealed: a small body of the uterus in anteflexion; macroscopically normal left adnexa; in the right adnexa – an ovarian tumor (7 x 9 cm), a cyst in the mesosalpinx (2 x 3 cm). Following a pathological examination the diagnosis of a benign teratoma (adultum cysticum) was confirmed. [Figure 1] Macroscopically: encapsulated, dissected tumor (6 x 6.5 cm), with sebum and hair seen on the cross-section. In the mesosalpinx, a paraovarian cyst was found. The Ca 19.9 level was 169.9 U/mL at two weeks post-surgery, and returned to normal values after a month.

**Figure 1 : The microscopic image of the mature cystic teratoma tissue.**

**DISCUSSION**

In 10% of cases dermoid cysts can occur bilaterally, and if unilateral, they are found more frequently on the right (60 - 70%) [3, 5]. To date, several cases of a marked increase in the Ca 19.9 level, similar to our case, have been described. Atabekoglu et al. reported a case of a 25-year-old symptomatic girl (virgo) with a lesion (30 cm in diameter) in the right adnexa, which was found on an ultrasound scan and confirmed in CT [6]. The imaging studies suggested a dermoid cyst. The level of Ca 19.9 was elevated up to 1430 U/mL, and the level of Ca 125 was 46.6 U/mL. The lesion was confirmed to be a dermoid cyst. The authors suggested that Ca 19.9 is secreted by the endothelium found inside the cyst.

Therefore, the concentration of this marker increases quickly following a rupture or if an inflammatory reaction takes place inside the cyst. This can cause an increased permeability of the tissue resulting in glycoprotein Ca 19.9 moving from the cystic cavity into the systemic circulation. The larger the diameter of the cyst, the thinner its wall, and the greater the outflow of Ca 19.9. Uğur et al. described a correlation between an increase in Ca 19.9 level and the cyst size, the presence of adhesions in the abdominal cavity and an elevated level of Ca 125 [7]. Coscu et al. studied 43 patients with dermoid cysts. The level of Ca 19.9 in 39 cases was associated with the diameter and the weight of the tumor [5]. In our case, the tumor diameter was 6 cm. Only Mikuni et al. did not find any correlation between the weight and diameter of a cyst and an increase in Ca 19.9 level [8]. Dede et al. underlined that in mature teratomas an increase in Ca 19.9 was the most frequently observed anomaly (38.8% of cases), and was significantly more common in patients with bilateral tumors [4].

Kataoka et al. also underlined that the level of Ca 19.9 was significantly higher in patients with bilateral lesions, with no
difference between pre- and postmenopausal patients [9]. The highest observed concentration of Ca 19.9 in a benign dermoid cyst was 25590 U/ml. [Table 1] In our case, Ca 19.9 was non-specifically elevated. That elevation was not correlated with bilateral location, the tumor was not large, and the lesion was pathologically confirmed to be benign. Other authors reported that the level of Ca 19.9 was increased in teratomas with a malignant component, but the values of Ca 19.9 never exceeded 1000 U/mL. Notably, torsion of a dermoid cyst caused a greater increase in Ca 19.9 [3]. Such a torsion causes ischemia with a subsequent increase in permeability of the cystic wall. It was observed that the level of the marker returned to normal values after circa 3 to 4 weeks [9, 10]. Dermoid cysts resulting in an increase in Ca 19.9 were also observed in extra-ovarian locations. Kahraman et al. reported a case of a 43-year-old patient with a lesion (75x58 mm) in the pouch of Douglas [11]. It was not possible to precisely locate the lesion during an ultrasound examination. Ca 19.9 and Ca 125 values were 1950 U/mL and 52.5 U/mL, respectively. Because of the large size of the lesion and an increased level of Ca 19.9 laparotomy was performed, during which a tumor in the sacral area was found. A pathological examination confirmed the diagnosis of a mature teratoma. Other authors described a dermoid cyst (5x3 cm) in the inguinal region, in the round ligament of uterus, which presented as an inguinal hernia [12]. In our case, in addition to the dermoid cyst, we found a pituitary tumor (prolactinoma). There are reports of prolactin being secreted by the glandular part of the pituitary located in the wall of an ovarian dermoid cyst [13]. Another case was a patient with galactorrhea, amenorrhea, biochemically confirmed hyperprolactinemia, in whom a prolactin-secreting dermoid cyst in the pituitary gland was found [14]. In our patient, a lesion suggestive of a hormone-secreting tumor (prolactinoma) was found on a head MRI scan, which prompted us to initiate treatment with dopamine agonists.

<table>
<thead>
<tr>
<th>Author</th>
<th>Average patient age [years]</th>
<th>Bilaterals [%]</th>
<th>Cysts diameter [cm]</th>
<th>Ca19-9 level [u/ml]</th>
<th>Surgery</th>
<th>Histopathology</th>
</tr>
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<tbody>
<tr>
<td>Ustunyurt et al [14]</td>
<td>36</td>
<td>9.8</td>
<td>8.8 ±4.5</td>
<td>83.5±179.2</td>
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<td>teratomaadultum</td>
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<td>Nagata et al [10]</td>
<td>50</td>
<td>left side only</td>
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<td>794</td>
<td>laparotomy</td>
<td>cystic teratoma</td>
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<td>Nanayakkara et al [12]</td>
<td>54</td>
<td>right side only</td>
<td>8x6</td>
<td>436</td>
<td>laparotomy</td>
<td>dermoidcyst</td>
</tr>
<tr>
<td>Ulkumen et al [3]</td>
<td>14</td>
<td>right side only</td>
<td>11</td>
<td>1983</td>
<td>laparotomy, right side adnexectomy</td>
<td>dermoidcyst</td>
</tr>
<tr>
<td>Kataoka et al [9]</td>
<td>32</td>
<td>17.2</td>
<td>41% &gt; 10</td>
<td>25% &gt; 500 highest 25590</td>
<td>-</td>
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<tr>
<td>Cho HY et al [15]</td>
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<td>15.45</td>
<td>8.5±3.84</td>
<td>114.6±20.66</td>
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<td>43</td>
<td>-</td>
<td>7.5x5.8</td>
<td>1950</td>
<td>laparotomy</td>
<td>dermoidcyst</td>
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<td>Ugur et al [7]</td>
<td>38</td>
<td>10.8</td>
<td>-</td>
<td>46.95±101.11</td>
<td>6.8% torsion</td>
<td>mature cystic teratoma</td>
</tr>
<tr>
<td>Coskun et al [5]</td>
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<td>7 (72% right side only)</td>
<td>-</td>
<td>-</td>
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<td>mature cystic teratoma</td>
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<tr>
<td>Dede et al [4]</td>
<td>34</td>
<td>27.5</td>
<td>7.2±4.5</td>
<td>101.2±179.7</td>
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<tr>
<td>Atabekoğlu et al [6]</td>
<td>25</td>
<td>right side only</td>
<td>30</td>
<td>1430</td>
<td>laparotomy, right side adnexectomy</td>
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<td>Kyung et al [15]</td>
<td>32</td>
<td>20.2 (47.9% right side only)</td>
<td>7</td>
<td>22 (4–575)</td>
<td>12.9% torsion</td>
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</table>

Table 1 : Comparison of cyst diameter, the level of Ca 19.9 and the type of surgery – review of the literature.
CONCLUSION

It seems that an isolated increase in the non-specific marker Ca 19.9 is rarely associated with a malignant transformation. Therefore, we recommend that the excision of dermoid cysts is performed laparoscopically regardless of the marker level, especially in young patients.

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REFERENCES


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