CASE REPORT

Borderline ovarian cyst treated by laparoscopic surgery: clinical case report and literature review

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Abstract
Borderline ovarian serous tumors are a rare distinct category of epithelial ovarian tumors, distinguished from both benign and invasive ovarian tumors. As borderline ovarian cysts are only a small part of the gynecological conditions of children, the therapeutic approach to this special type of tumor has not been standardized yet. Despite the technological breakthroughs that we have witnessed lately, the positive diagnosis of ovarian tumors is possible only by surgery and histopathological examination, and laparoscopic surgery has become the golden standard in the management of this condition. The aim of this case report is to demonstrate a very good long-term evolution after minimal invasive treatment and minimal damage to the reproductive apparatus in children with borderline ovarian tumors. The case reported in this paper is that of a 17-year-old adolescent with borderline serous papilliferous cystadenoma diagnosed by ultrasound and computed tomography (CT) scanning, whose CA-125 biomarker had normal values. As the purpose was to preserve the patient’s reproductive function, we chose to remove and excise the cyst by laparoscopic surgery, without removing the ovaries. The patient’s subsequent evolution was positive and at 19-year-old, she gave natural birth to a healthy baby girl. Nevertheless, as relapses are possible after many years, the patient’s long-term monitoring is necessary.

Keywords: borderline ovarian tumor, children, laparoscopy.

Introduction
Pediatric gynecological pathology is dominated by adnexal tumors, more precisely ovarian tumors. The incidence of ovarian tumors increases with age and it is considerably higher in girls over 14 years of age. Abdominal pain is the most common symptom, and the diagnosis of ovarian tumor is usually set by abdominal ultrasound scanning. Alpha-fetoprotein and CA-125 are the most frequently used biomarkers for diagnosis setting. The surgical treatment should preserve both the contralateral and the affected ovary in all cases, except when a malignancy is strongly suspected [5].

The case reported in this paper is that of a 17-year-old adolescent with borderline serous papilliferous cystadenoma. As the purpose was to preserve the patient’s reproductive function, we chose to remove and excise the cyst by laparoscopic surgery, without removing the ovaries. The patient’s subsequent evolution was positive.

Case report
This paper reports the case of a 17-year-old adolescent who is hospitalized for abdominal pains the onset of which occurred three days before and for right chronic otorrhea. She had no family or personal pathological medical history. The physical examination reveals a round mobile relatively well-delimited and slightly painful on palpation tumor located in the hypogastrium. The ENT (ear, nose and throat) examination sets the diagnosis of right chronic otomastoiditis and recommends antibiotic therapy and cranio-cerebral computed tomography (CT) scan designed to set the subsequent therapeutic approach. The biological tests including inflammatory markers, tumor markers and chest and abdomen X-ray have all normal values. The CA-125 biomarker had normal values before surgery (28.15 IU/mL), as well as after six months and one year (28.15 IU/mL). The abdomen ultrasound scan reveals a 14/8.5 cm big well-delimited liquid mass, with floating...
hyper-reflective sediment inside, located in front of the uterus and on top of the urinary bladder; the scan reveals many nodular budding images of the posterior and lateral wall (Figure 1). The left ovary exhibits several follicular cysts, the biggest having 2.3/1.8 cm. The native and contrast medium abdomen–pelvis CT scan detects a 9.8/12.6/13 cm big well-delimited liquid cyst, the density of which ranges from 4 to 22 UH, located in the pelvis and belonging to the right ovary. The inner anterior and inferior wall of the cyst is lined with several solid masses of sessile appearance and wide implantation bases, with contrast uptake after contrast medium injection, the biggest of which have 1.6/0.9 cm and 1.1/2.3 cm. No pelvic adenopathies are revealed (Figure 2). The cranio-cerebral CT scan exam with contrast medium reveals right chronic otomastoiditis and nasal septum deviated to the right.

A laparoscopic surgical procedure is performed, the peritoneal cavity is explored and the well-delimited tumor is observed (Figure 3). It has no adherence to the neighboring organs and it belongs to the right ovary. The tumor is considered to be in the FIGO stage I [6]. A puncture is performed for tumor removal and then cyst enucleation and excision (Figure 4). Considering the normally looking pelvis and absence of adenopathies, as well as the patient’s age and wish to have children, both ovaries are preserved. After the surgery, she was given antibiotic therapy (also for the ENT condition) and support therapy, and as her evolution was positive, she was discharged three days after the surgical procedure. A year after the operation, the patient’s evolution was positive, the biological tests had normal results, and the abdomen ultrasound scan revealed normally looking pelvis and adnexa. At 19-year-old, she gave natural birth to a healthy baby girl, 3250 g, 40 weeks gestational age. During pregnancy, the patient was monitored serologically for tumoral markers (inflammatory markers, alpha-fetoprotein, CA-125) and with repeated ultrasounds and all the tests were in normal range.

The histopathological examination performed in order to determine the exact clinicopathological entity of the giant tumor revealed on the analyzed specimens ovarian stroma with many connective–vascular axes lined by a highly prismatic epithelium, stratified here and there (Figures 5–7). Some axes exhibit fibro-connective proliferation, others are long, have filiform appearance and reduced stroma. The cells of the epithelium lining the papillae exhibit hyperchromasia. The proliferating cells have minimally and moderately atypical nuclei. Scarce mitosis, calcifications and areas of hemorrhage and vascular stasis are also revealed. The proliferation cells do not invade the underlying stroma (Figures 8–10). We performed immunohistochemistry using EnVision Flex-DAB staining system and the monoclonal antibodies used were anti-CD45, anti-CD68 and Ki67 proliferation factor. The examination revealed minimum inflammation inside the cyst wall (few T- and B-lymphocytes – CD45 and macrophages – CD68). The Ki67 proliferation index is high (20–30%) on some papillary projections and low (10%) on the rest of the epithelium (calculated on 10 high-power fields ×400) (Figures 11–13). Examination result: borderline serous papilliferous cystadenoma (low malignant potential tumor), according to the World Health Organization (WHO) classification of ovarian tumors.
Figure 5 – Cyst wall (ovarian stroma with many connective-vascular axes) lined by a highly prismatic epithelium, stratified here and there. Szekely (Sz) trichromic staining, ×100.

Figure 6 – Cyst wall with a tendency to develop projections. Sz staining, ×200.

Figure 7 – Cyst wall: fibrous tissue and simple cubic epithelium inside. HE staining, ×100.

Figure 8 – Papillary projections on the anterior inside wall of the cyst, with connective-vascular axes, some with collagen, others with cellularization. HE staining, ×40.

Figure 9 – Papillary projections with vascular axes and calcification. HE staining, ×200.

Figure 10 – Papillary projections: the proliferating cells have minimally and moderately atypical nuclei and exhibit hyperchromasia. HE staining, ×200.
Discussion

Borderline ovarian tumors are on the border between benign cystadenoma and invasive cystadenocarcinoma, as they have histological and cytological malignancy characteristics yet without stroma invasion, and they make up 10–15% of all epithelial ovarian tumors [7]. Serous borderline tumors are characterized by moderate or marked proliferation of the neoplastic cells, cell stratification and papillae formation inside the cyst. Here are their morphological characteristics: epithelial cell stratification, high mitotic activity, nuclear abnormalities, cell atypias. Neoplastic cells usually have a small amount of cytoplasm, but sometimes they may be big, with a high amount of eosinophilic cytoplasm, although this type of cells rarely predominates. There may be variable degrees of nuclear atypias, the mitotic aspects ranging from rare to numerous. The histological criteria remain the only method of distinction between borderline tumors and invasive ovarian carcinomas [8]. On our case, the Ki67 proliferation index was high (20–30%) on some papillary projection and these nests could be the starting point of a malignant proliferation.

Even borderline tumors may sometimes lead to death by metastatic illness, although the progression and metastatization are slow. The overall five-year survival rate of patients with serous and mucinous borderline tumors generally ranges between 74% and 98% [9]. In any case, the survival rates for the 1st stage of borderline tumors are excellent, being close to 100%, and significantly better than for invasive carcinoma. Serous borderline tumors make up 5–15% of all ovarian serous carcinomas. Macroscopically speaking, most of them are cystic and papillary, whereas from a histological point of view, they are non-invasive neoplasms characterized by multiple fibrous papillae covered by a multilayered epithelium with cell exfoliation and mitotic appearance. Borderline serous tumors often secrete a thick mucinous liquid and a differential diagnosis with mucinous tumors is then necessary. In the 1st stage, up to 25% of them are bilateral, and in other 10% of the cases the contralateral tumor is revealed only by a microscopic exam [10]. According to other studies, bilateral tumors occur in up to 65% of the cases [11]. The recurrence rate may be of up to 44% within the 15 years following the first occurrence, and about 10% of the patients in whom the other ovary was preserved developed a second borderline primary tumor in the remaining contralateral ovary. From the histological point of view, relapses are similar to the primary tumor and even regressions, though rare, have been reported [12, 13]. The highest number of cases occur in patients aged between 30 and 60 years of age [14], the condition being very rare in patients younger than 18, like our patient. A study on 244 ovarian masses in 219 pediatric patients identified only four (1.6%) borderline malignant lesions (borderline epithelial tumors) [15]. By a basic preoperative evaluation, the risk of unsuspected borderline or focally invasive ovarian cancer significantly increases by age, size, complex morphology and CA-125 [16].

The most adequate therapy for low malignity tumors has not been set yet; it seems that in the 1st and 2nd stages of the disease, surgical resection alone has excellent results, therefore the therapy of 1st stage tumors occurring in young patients who want children is unilateral adnexectomy without adjuvant therapy, and close clinical and biological follow-up [13, 17]. Laparoscopic treatment of adnexal masses has proven to be a safe and effective diagnostic and therapeutic tool in the hands of experienced laparoscopists, especially in young patients [17].
is an increased risk of perforation or rupture with trocar insertion and excision during laparoscopy for treatment of giant ovarian cysts. Also, these cysts may exceed the working space [18]. Mayer et al. suggested that laparoscopy for treatment of ovarian cysts, including those with large dimensions, is associated with satisfactory outcomes and few complications; the giant cyst can be punctured and the cyst wall subsequently removed [19]. Laparoscopy is preferred over open surgery for the treatment of ovarian masses in children and adolescents due to shorter surgery duration, blood loss and less hospitalization days [20]. A recent study demonstrated that even when laparoscopic surgery is used for the resection of an adnexal mass that is most likely benign, some patients are found to have malignant tumors post-operatively. The rate of unexpected ovarian malignancy resected by laparoscopy was 1.5%, but the presence of an early-stage unexpected ovarian malignancy did not alter patient prognosis [21]. However, with careful patient selection, proper technique and an experienced operator, laparoscopic treatment of ovarian masses is the best approach and should be implemented [22]. Considering our patient’s age, her first stage tumor and her clear desire to have children, we performed exploratory laparoscopy to view the tumor. As the macroscopic examination supported its benignity and revealed no adenopathies, we decided to remove only the cyst and preserve the ovaries, the subsequent evolution of the patient being extremely positive. Retrospectively, seems like it was a good decision, because the patient actually gave birth to a healthy baby, one year and a half later. Unlike ovarian cancer confirmed by histological examination, in borderline tumors the systematic dissection of the lymphatic nodes is not necessary [23, 24]. The CA-125 antigen had normal values in our case, but it is generally high in patients in advanced stages of the illness. Higher values are more frequently detected in the serious rather than in the mucinous histological diagnosis [25]. Considering the possibility, although rare, of later relapses or of occurrence of a similar tumor on the contralateral ovary, the patient will require long-term clinical, biological and imagery monitoring. Clinical follow-up remains the gold standard for benign ovarian tumors, and it seems that 6-month laparoscopic look is a better strategy for monitoring patients with high-risk borderline ovarian tumor than ultrasound/CA-125 evaluation [26].

Conclusions

Over the last decades, the management of borderline ovarian tumors has changed from radical and mutilating surgery to more conservative therapy because of the need for fertility-sparing surgery and the increasing use of laparoscopy. We demonstrated a very good evolution after minimal invasive treatment and minimal damage to the reproductive apparatus in a young girl with borderline ovarian tumor, which only one year and a half later gave natural birth to a healthy baby. Considering the possibility, although rare, of later relapses or of occurrence of a similar tumor on the contralateral ovary, the patient will require long-term clinical, biological and imagery monitoring.

Conflict of interests

The authors declare that they have no conflict of interests.

Author contribution

All authors have equally contributed in preparing this manuscript and thus share first authorship.

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For publication of this article, the written informed consent of the patient and her mother (her father being died) was given.

References


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