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Ovarian Teratoma In Pediatric Population: Our Clinical Experience of Seven Years

Çocuklarda Ovaryan Teratomlar: 7 Yıllık Deneyimimiz

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ABSTRACT

Objective: In relation to torsion, mass appearance may occur due to hemorrhage into the cyst, and blood supply of the over tissue may not be well evaluated radiologically. For this reason, the diagnosis of torsion and non-torsion MCT continues to be a serious dilemma.
Material and Methods: The laboratory and imaging methods of ovarian teratomas with or without torsion were compared with the statistical method. The results were evaluated statistically by using SPSS version 24. Normality checks were performed with the Shapiro-Wilk test. The groups were compared with the independent samples t-test. Statistical significance was p <0.05.
Results: 40% of the cases with CRP level elevation, torsion were significantly higher (> 5 mg / L). The most common surgical approach was fertilization protective cystectomy applied in 14 cases (77 %).
Conclusıon: The first choice in benign ovarian masses is fertility protective surgery. CRP, WBC and CA19-9 can be a marker, especially in torsion cases. The planning of the study with prospective and large number of patients will be appropriate.

Key Words: Mature cystic teratoma, ovarian torsion, pediatrics

ÖZET

Amaç: Torsiyon ile ilişkili olarak kist içine kanama nedeniyle kitle görünümü oluşabilmektedir ve over dokusunun kanlanması radyolojik olarak net değerlendirilemeyebilir. Bu nedenle torsiyone ve torsiyone olmayan MKT' lerin tanısı ciddi bir ikilem olmaya devam etmektedir.
Gereç ve Yöntem: CRP yüksekliği torsiyonun eşlik etiği vakaların %40'ında anlamlı derecede yüksek(>5mg/l) bulundu. En sık yapılan cerrahi, 14 vakada uygulanan fertilite koruyucu kistektomi (%77) idi.
Bulgular: Torsiyon olan ve olmayan ovaryan teratomların laboratuvar ve görüntüleme yöntemleri istatistiksel yöntemle karşılaştırıldı. Sonuçlar SPSS versiyon 24 kullanılarak istatistiksel olarak değerlendirildi. Normallik kontrolleri Shapiro-Wilk testi ile yapıldı. Gruplar bağımsız örneklem t-testi ile karşılaştırıldı. İstatistiksel anlamlılık p <0.05 idi.
Sonuç: Prospektif ve çok sayıda hasta ile çalışmanın planlanması uygun olacaktır. Benign ovaryan kitlelerde ilk tercih fertilite koruyucu cerrahidir. CRP, WBC ve CA19-9 özellikle torsiyon vakalarında işaretleyici olabilir.

Anahtar Sözcükler: Matür kistik teratom, ovarian torsiyon, çocuk

Introduction

Ovarian masses are quite rare in children and the incidence is 2.6 in 100 000. Clinical presentation and pathology are quite different from adults (1). Ovarian masses can be classified as neoplastic and non-neoplastic. Teratomas are the most frequent ovarian masses in children (2). And they are the most common benign tumours of ovary (2,3). The most frequent histopathologic type of ovarian teratomas seen in the pediatric population is mature cystic teratoma (MCT).

It can be detected with an acute abdomen or coincidentally during routine examination. In relation to torsion, mass appearance may occur due to haemorrhage into the cyst, and blood supply of the over tissue may not be well evaluated radiologically. This may lead to elective planning of the surgical procedure and delay of the detorsion procedure that must be performed in an emergency. Ovarian torsion with teratoma results in a decrease of healthy ovary tissue. Also; the appearance of mass during surgical interventions due to acute abdomen may cause unnecessary oophorectomies because of the suspicion of malignancy in the surgeon. The combination of teratoma and torsion is a diagnostic dilemma.

This study was retrospectively evaluated in terms of age, clinical presentation, diagnostic and imaging

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Fig. 1. MCT with torsion (left ovary)

methods, additional pathology, surgical technique and histopathologic results in patients who underwent treatment for ovarian teratoma in our clinic.

Materials and Methods

This work was carried out in accordance with the Helsinki Declaration principles. Also, by the local ethics committee for study approval received.

In this study, clinical application, examination findings, laboratory results of 0-15-year-old girls who were pathologically diagnosed as MCT in our clinics between January 2010 and January 2018, imaging techniques, surgical techniques, complications and recurrence were retrospectively evaluated. Those who were unable to reach clinical data, those with additional ovarian pathology, and patients older than 18 years were excluded from the study.

Patient information was accessed through the hospital registry system. Hemoglobin, hematocrit, platelet count, white blood cell count and C-reactive protein (CRP) values were documented from the laboratory findings. Results of ca-125, ca 19-9 and alpha-fetoprotein (AFP) values were compared as tumor markers. The imaging modalities and radiological reports of patients were evaluated. In these cases, the laboratory and imaging methods of ovarian teratomas with or without torsion were compared with the statistical method. The results were evaluated statistically by using SPSS version 24. Normality checks were performed with the Shapiro-Wilk test. The groups were compared with the independent samples t-test. Statistical significance was $p < 0.05$.

Results

18 female patients with a median age of 13 years were included in the study. Nine patients had no symptoms at first visit. Abdominal pain in 5 patients, mass in 2 patients, fever in 2 patients, vomiting and loss of appetite in 5 patients and abdominal fullness were present in 3 patients. Laboratory findings showed that

Fig. 2. Huge MCT with torsion (right ovary)

WBC elevation for their age scale was detected in 22% of patients and no significant difference was found between teratoma case with or without torsion.

Anemia was detected in 18% of patients. CRP level elevation was significantly higher (> 5mg / l) in 40% of teratoma cases with torsion. Patients were treated with different surgical methods. The most common surgical approach was fertilization protective cystectomy applied in 14 cases (77%). Salpingoopherectomy was performed in 1 case (5.5%), oophorectomy in 3 cases (16%), and ovarian biopsy in 9 cases (50%). Oophorectomy was performed mainly because of suspected malignancy and ischemia. 21 overcysts were excised from a total of 18 patients (bilateal mass in three patients).Four of the ovarian masses were excised from left, and 17 of them from the right over. Intraoperative torsion (figure1-2) was detected in 5 patients (27%). Hemorrhage was seen in all of the teratoma cases with torsion and necrosis in one. The mean mass size was 12 cm and it was calculated as 5.4 cm in the teratoma cases with torsion.

In one of the ovarian teratoma cases acute appendicitis also was seen co-incidentally. No recurrence was detected during follow-ups. Tumor markers were also checked in 9 of the patients (50%). CA-125, CA 19-9, and AFP level were checked on 1/5/9 of them respectively. CA 19-9 was high in 2 patients. AFP levels were high in 3 patients (> 40 ng /mL). Ca 19-9 level of the teratoma cases with

Table 1. Laboratory Results, Performed Imaging Methods and Applied Surgical Methods İn MCT Cases With Or Without Torsion

	MCT	MCT with Torsion	p
	Labaratuary		
Hb	12	11,3	P=0,06
Htc	39	36	P=0,07
WBC	11.000	12.000	P=0,06
CRP	3,1	45	P=0,02
AFP	48	12	P=0,03
CA 19-9	<0,2	88	P=0,04
CEA 125	20	23	P=0,08
	Radiology		
Ultrasound	13	5	P=0,05
CT	4	0	
MRI	1	0	
	Surgical Approach		
Conservative Surgery	10	4	P=0,08
Oopherectomy	3	1	P=1
	Mass Size		
	12	5,4	P=0,01
Patients	13	5	18

torsion was higher than those of the non-torsion teratoma cases, but no significant difference was checked between the other tumor markers. It was evaluated according to imaging methods used for diagnosis in patients; ultrasound was used in all patients, MRI was used in one and CT was used in three. Ultrasonography was deemed adequate for surgical indication in 14 of the patients. In 2 cases, surgery was indicated with MRI, whereas in 2 cases, surgery was indicated with CT. When the surgical methods are examined; Open surgery was performed on the sixteenth patients and laparoscopic surgery was applied in two patients. In MCT patients with or without torsion; doppler ultrasonography performed at the sixth month after ovarian conservative surgery revealed that the amount of healthy ovarian tissue and blood supply were similar (table). No malignancy was found in the pathology specimens of these 18 patients.

Discussion

Approximately one third of adnexal masses are ovarian tumors (2). Small ovarian cysts can be detected incidentally in prepubertal children 2-5%, especially during neonatal period due to maternal and placental hormonal stimulation (4) and these benign lesions often disappear without treatment. Over teratomas, which are the most common germ cell tumors, are surgically removed. Teratomas contain numerous histological types of tissue based on mature and immature germ cell (pluripotent) origin. The most common type of these tumors, MCT (also known as dermoid cyst), typically consists of mature ectodermal tissue (skin, brain), mesodermal tissue (muscle, fat) and endodermal tissue (mucinous, ciliated epithelium) (3,5). The incidence of MCT is 1.2-14.2 (7) at 100.000 and the incidence increases with age. It is most commonly seen around 14 years (2). MCT constitutes 25% of all ovarian tumors and 60% of benign ovarian tumors (6,7). Bilaterality probability is 10-15% (3). Unilaterality is mostly the right adnexal location (6). 60% of them is about 5-10 cm in size. Tumor size is unrelated to malignancy potential (4). Abdominal pain and abdominal fullness are the most common symptoms (2) but may also be detected incidentally. Rupture; despite the thin wall of the cyst is quite rare. Torsion is detected at 15% of the MCTs. The incidence of torsion increases in cysts with long pedicle and 5-6 cm size (8).

Radiologically; , USG, CT and MRI diagnose pelvic pathologies in 77%, 87% and 97%, respectively (4). Ultrasonography is the gold standard modality for diagnosis of MCT (2,5). However, it is very difficult to reveal the torsion with sonographic examination (6). radiologically; Ovarian haemorrhage may cause complex and solid appearance (4) and this creates a diagnostic dilemma.

Tumor markers are frequently used to identify tumor etiology. AFP and CA 125 are the most commonly used tumor markers (2). CA 125 and CA 19-9 levels are significantly elevated in patients with squamous carcinoma associated with dermoid cysts. CA-19-9 levels correlate with tumor size. The increase of Ca19-9 level in serum was due to mass necrosis; CA 125 is elevated in gastric irritation caused by escape of the cyst fluid to the peritoneal cavity and in pelvic inflammatory disease (6). CA 125, CA 15-3 and AFP can be useful in differentiating MCT and immature teratoma (IT) cases (5). As in our cases; Elevation of CRP, CA 19-9 and leucocyte levels may be a marker for dermoid cyst cases with torsion.

If the cystic mass is suspected of malignancy because of the complex and solid components, if there is intermittent or continuous abdominal pain in the patient, or if there is a simple cyst greater than 5 cm, surgery is indicated (4). Overian cystectomy is the gold standard, unless there is a doubt about malignancy in young patients (4,8). Over the first year after over-conservative surgery, 70% of the healthy ovarian tissue is protected more than 3 cm^3 (8).

Laparoscopy is the gold standard in MCT surgery (2).With laparoscopy, the amount of intraoperative bleeding is decreased, the postoperative pain is relatively less, the length of hospitalization is shortened, the risk of postoperative adhesions is decreased and good cosmetic appearance is achieved. However, it has disadvantages such as prolongation of the surgical intervention, leaking of ruptured cystic tissue into the abdomen, and high recurrence rates [8]. The rate of leakage of cyst fluid into the abdomen during laparoscopy is reported to be approximately 100%, but the formation of chemical peritonitis associated with fluid is rare (0.2%) (2).

Limited of our study, the number of patients is not sufficient to statistically indicate the outcome. The planning of the study with prospective and large number of patients will be appropriate.

Ovarian masses in children and young adolescents are often benign teratomas. The first choice approach in children with benign ovarian masses is fertility protective surgery. Changes in levels of CRP, WBC, and CA19-9 can be a marker, especially in teratomas with torsion.

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