CME LIVE OPEN ACCESS

Journal of
Gynecology and Reproductive Health

Research Article

Mature Ovarian Teratoma In A 13-Year-Old Girl

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Received: 2024 May 09 **Accepted:** 2024 May 29 **Published:** 2024 June 01

Abstract

Ovarian tumors represent 1% of childhood tumors. In children, they are often benign and can recur, but the future fertility preservation is essential. Ovarian teratomas are the most common ovarian germ cell tumors. A bimodal distribution is observed with a first peak in early childhood and a second peak after puberty. During childhood, the location is mainly sacrococcygeal and gonadal in adolescence. These teratomas can be mature and benign: either polytissue containing ectodermal derivatives (hair, teeth, sebum), which are dermoid cysts; or mono-tissue, containing thyroid tissue for example (ovarian goiter). As for immature and malignant teratomas such as endodermal sinus tumors, they secrete alpha-feto-protein, while ovarian choriocarcinoma secretes HCG. We report the illustrated observation of a mature ovarian teratoma case in a 13-year-old patient at Djibouti military hospital. The adolescent was suspected of being pregnant by some of her parents and the clinical examination revealed an abdominal-pelvic mass with compression signs. CA125 was very high, alpha-fetoprotein and BHCG negative. Ultrasound and especially CT were in a large ovarian teratoma favor. The surgical procedure was an adnexectomy; anatomy pathology confirmed a mature ovarian teratoma and the sequels were simple.

1. Introduction

The ovary is a complex organ housing many different cell types: germ line cells and theca cells, stromal cells (connective tissue) and epithelial cells of the mesothelium covering the surface of the ovary. This tissue richness explains the great variety of tumors on the histological, physiopathological and prognostic levels [1]. Ovarian tumors are one of the most difficult problems in gynecological pathology in terms of clinic, histology and prognosis, given the polymorphism they present [2]. Ovarian teratomas are detected in 37.5% of germ cell tumors [3]. They are the most common type of tumor in children. However, some aspects of its pathology, classification and management still remain unclear [4].

Dermoid cysts or mature teratomas arise from a multipotent cell that may be the origin of different tissues present within the cyst (adipose tissue, body hair, bone, tooth, etc.) [1]. On the macroscopic anatomical level, the teratoma can be uni or multilocular, divided by septa into a number of compartments. The cystic component is generally represented by a fatty liquid, made of sebum, keratin and hair surrounded by a firm capsule. This content is usually in a liquid state at temperature above 34°C. Fat in the cyst lumen is present in more than 93% of cases and it is the most specific imaging finding indicative of teratoma cystic mature. The lipid content of mature cystic teratomas is largely sebaceous fluid and more rarely adipose tissue [4].

Histologically, mature cystic teratoma presents a variable mixture of elements from one or more of the three cell lineages: derived from ectodermal, endodermal, mesodermal tissues and the lesion is bilateral in 12% of cases [4]. Ectodermal tissue (cutaneous derivatives and neural tissue) is invariably present; mesodermal tissue (bone, fat,

cartilage and muscle) is present in more than 90% of cases, and endodermal tissue (thyroid, gastrointestinal tissue) is present in the majority of cases [4]. Malignant (immature) transformation of one or more of the components of the teratoma is rare, affecting 1 to 3% of mature cystic teratomas [5]. The circumstances of discovery vary, the clinic is not easy, especially in children and adolescents, but the diagnosis must always lead to intervention. Imaging, hormonal dosage as well as tumor markers and anatomical pathology are an important contribution to the management and prognosis of these tumors.

2. Our Observation

SKH is 13 years old, she is the fifth child of 09 siblings, with no known particular personal and family history. She is single, nulligest, normal regulated since her menarche at 11 years old. She reported to us that she had never had sexual contact, had been experiencing episodes of nausea and heaviness-like abdominal-pelvic pain for several weeks and was on day 7 of the menstrual cycle at admission. Accompanied by her father, she was admitted to the department for suspicion of pregnancy. On examination, the general condition was good, hemodynamic constants without particularities, female morphotype, breasts and hair at stage 4 of Marshall and Tanner maturity scale. We found a large, painless, firm and mobile abdominopelvic mass measuring 18x13 mm (Figure 1).

Figure 1: Clinical Appearance
Abdominopelvic Mass, Patient in Supine Position (Black Arrow).

The vulva was clean, excision was notedtype II; the vaginal examination was not done and the hymen appeared intact. The rectal examination was not continued because of the discomfort and pain felt by the patient. The rest of the examination found shiny, painless edema with pitting on both legs and feet. At the end of this examination, we discussed three diagnostic hypotheses in order of priority: an ovarian tumor, an uterine fibroid and a pregnancy, then we began assessments. Abdominopelvic ultrasound revealed a large heterogeneous mass, independent of the uterus but difficult to identify; the uterus and right ovary were normal in appearance (Figure 2).

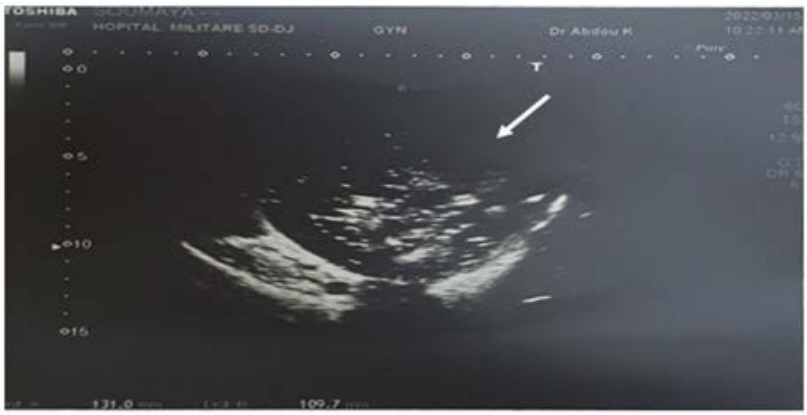

Figure 2: Abdominopelvic US
Heterogeneous mixed mass, thick wall (white arrow)

Abdominopelvic CT revealed a median abdominopelvic cystic formation measuring 17x11x9mm containing fine septa, calcifications and fatty tissue suggesting a mature ovarian teratoma, without ascites and without other notable abnormalities (Figure 3).

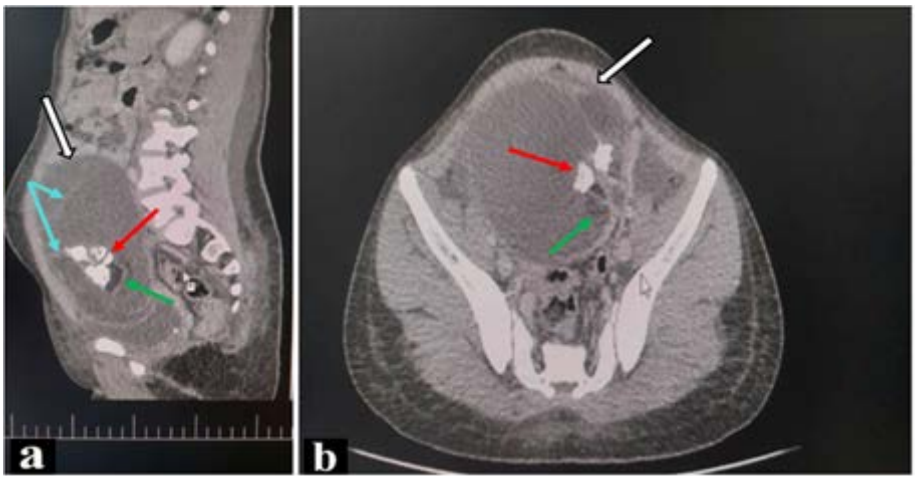

Figure 3: Abdominopelvic CT
In Sagittal (a) and Axial (b) Sections: Tumor (⟹) with Partitions (→), Calcification (→) and Fatty Tissue (→)

BHCG was negative and CA 125 elevated to 154.5U/l (more than 4 times normal). We discussed with the patient and her family and obtained their agreement for surgical intervention. The preoperative assessment and anesthesia consultation were unremarkable. At midline subumbilical laparotomy, we found this large cystic mass affecting the entire left ovary without any apparent distinct healthy tissue, the tubes without particularity, the contralateral ovary slightly enlarged and cystic; the peritoneum and other accessible intra-abdominal organs were also visibly normal in appearance, without ascites or other pathological effusion (Figure 4).

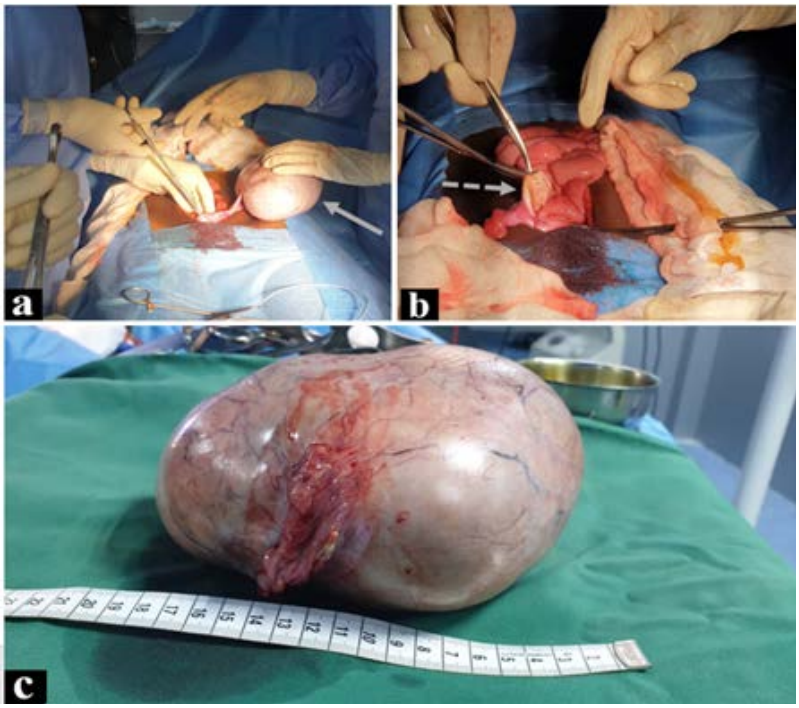

Figure 4: Per and Post Laparotomy Pictures
a- Left Ovarian Mass (⟶). b-Right Ovary also Cystic, Biopsy after Drilling (- - ▸). c- Left Annexectomy Piece: Ovary with Tumor+Tube.

The main procedures performed were: a left adnexectomy, drilling (bringing back serous fluid) and a biopsy of the right ovary. The various parts were sent for anatomo-pathological examination and the postoperative course was simple. On macroscopic examination the left adnexectomy piece weighed 1253 g; the left ovarian tumor, encapsulated, of renitent consistency and smooth whitish surface measured 16x13x12cm. On section, a unilocular cystic cavity with partitions was observed, containing sebomatous fluid associated with hair and teeth (Figure 5).

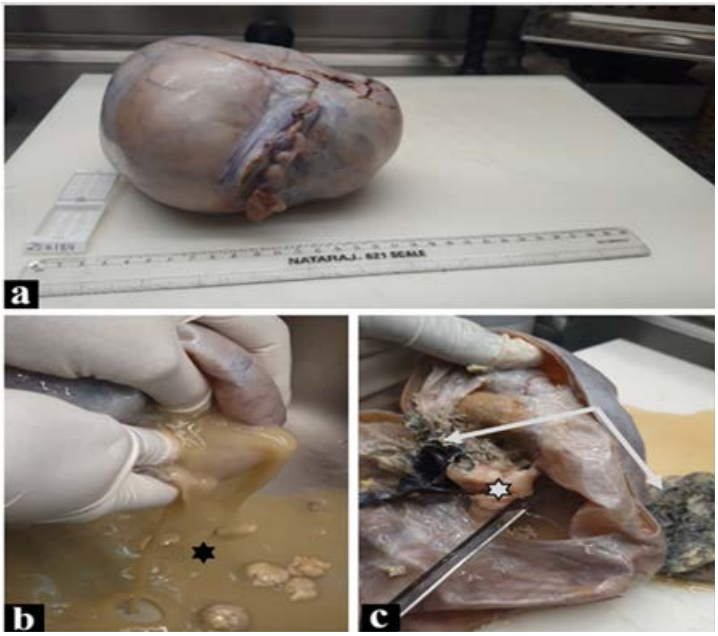

Figure 5: Macroscopic Anatomy
a- Tumor Measuring 16x13x12 cm with Well Encapsulated Surface.
b and c- Slice of Section of the Tumor with Sebum(✸), Teeth (✡) and Hair (⟶).

Microscopic examination of the histological sections of the left ovary revealed a cystic wall covered by a squamous epithelium which rests on a fibro-muscular tissue with foci of calcium changes and a moderate lymphocytic infiltrate, suggesting a teratoma. mature ovary (Figure 6). As for the right ovarian tissue, these were follicular cysts without signs of malignancy.

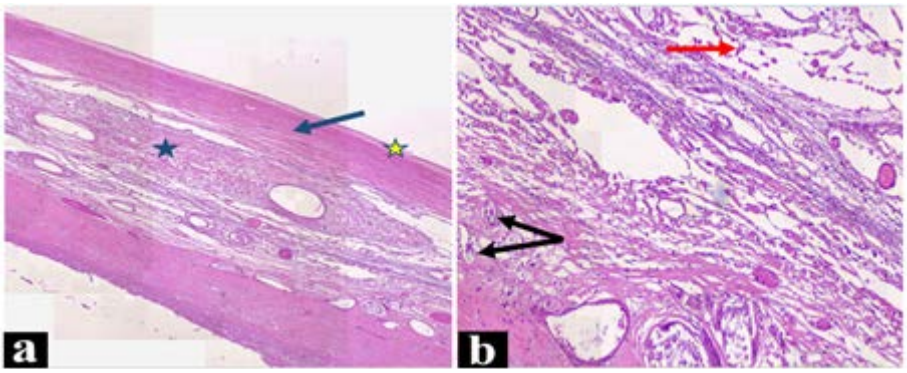

Figure 6: Microscopic Anatomy, Mature Teratoma
a- Cystic Wall (⟶), cOvered with a Squamous Epithelium (✭) Resting on a Fibrous and Muscular Tissue with Calcium Changes (★): Medium Magnification, Hematoxylin and eosin Staining. b- Cystic Wall Comprising Tissue Muscle (⟶), Calcium Rearrangementsand Multi-Nucleated Giant cells (⟶): High Magnification, Hematoxylin and Eosin Staining

3. Discussion points

From Diagnosis To Treatment

Age of Onset: Mature ovarian teratoma in children or adolescents has been documented by other authors: median age 11 years at first surgical intervention, by MC-Williamson team; 14 years in the study by Moyambe JNT et al, 12 years with a standard deviation of 4.2 for Łuczak J et al; 11.2 years in the study by Özcan R et al [4, 6, 7]. These results are superimposable to ours: 13 years at diagnosis. However, M. Terenziani's team included 219 patients in their study, including 150 cases of mature teratoma with a median age at diagnosis of 55 months [8, 9].

Clinical: The clinical diagnostic elements of an ovarian teratoma are varied: from almost nothing, through nausea, vomiting, pain and/or pelvic mass, to its accidental discovery during adnexal torsion. We summarize our case as a large firm abdominal-pelvic mass, edema of the lower limbs in a context of nausea and abdominal-pelvic pain in a 13-year-old girl, suspected by her parents of being pregnant. Our clinical history is similar to that of Moyambe et al in a case report published in 2021 in Lubumbashi; by Rousseau MC et al in a case report published in 2022 in Belgium and by Łuczak J and Bagłaj M in a retrospective study on 58 patients aged 0 to 18 years, published in 2018 in Poland where they

also described 02 cases of appendix torsion [3, 4, 7].

Imaging: Ultrasound is until now the most commonly used imaging modality for the ovarian pathologiesevaluation. It makes it possible to confirm the presence of the mass and determine the organ of origin as well as its characteristics and possibly to identify associated anomalies, such as ascites, other pathologies or organ lesions [10]. This examination was even the only preoperative imaging for the case of Moyambe JNT et al [7]. E Peroux et al in 2015 revealed in their study concerning 41 patients, a performance of 40 ultrasounds; 27 CT and 12 MRI. In their series, 04 patients were operated on the basis of sufficient ultrasound results (without CT or MRI). CT and MRI show excellent sensitivity of the characteristics of the tumor: identification of fat, fluid, partitions, calcifications which represent the most frequent diagnostic results of mature teratomas [10, 11]. In our history, apart from this large mixed mass, the other ultrasound information of the abdomen and pelvis as well as the site of this tumor were of little contribution, thus motivating us to add a CT scan.

Tumor markers: Mature teratomas, initially benign tumors, are generally non-secreting. However, the role of tumor markers (AFP, B-HCG, CA125, CA19.9, and LDH) in the discrimination between benign and malignant tumors is essential according to some authors but controversial according to others. They can indeed be confusing, because a benign ovarian tumor in children could be associated with an increase in serum levels reported in approximately 20% of cases [8]. Of the 58 patients in the study by Łuczak J and Bagłaj M, 55 had a mature teratoma but tumor markers were available in 38; positive in only 05 (CA 125 in 04, AFP in 01) and negative in 33 [4]. Also very high levels (5 times the normal) of CA 125 as in our case, and CA 19.9 were noted in the observation of Rousseau MC et al which was also a mature teratoma [3]. Also, biohumoral alterations were associated with 75% of mature teratomas and 25% of immature teratomas in the study by Spinelli C et al [8]. However, these serum markers have not been explored by certain authors either pre- or postoperatively [7, 9]. In our case, the CA 125 was very high: 154.5U/ml (norm: 0 to 35), the negative carcino-embryonic antigen: 0.54 ng/ml (< 5.09) and BHCG negative (< 2 mIU/ml).

Surgery: The problem of preserving ovarian tissue in cases of teratoma is at the center of the management of these tumors. Some authors recommend cystectomy and ovarian conservation, especially when the woman is young [1]. Thus, a study carried out and published on 10 years of data by Spinelli C et al in Italy in 2021 made it possible to compare a multicenter surgical experience on ovarian teratomas in children with current trends in management [8]. Concerning the retrospective study, one hundred and ten patients with a mean age at diagnosis of 11.8 years were included and teratomas represented 90% of mature cystic masses.

Surgically, 78 oophorectomies were performed, 32 interventions were able to preserve the ovarian tissue and laparoscopy took place in 16.3%. As for current management trends, the average age at diagnosis was 11.9 years and 80.5% of cases were represented by mature teratomas. Of the 430 interventions, 331 oophorectomies versus 99 ovarian conservation surgeries were performed, as well as laparoscopy in 23.8% of cases. Similar results were found by Gkrozou. F et al in 2022: their research included 40 patients with an average age of 11.8, 90% (n=36/40) of mature cystic teratoma and 10% (n=4/40) of teratoma immature [12].

These patients all underwent surgery; thus 75% benefited from an oophorectomy and 25% from ovarian conservation. Oophorectomy was also the standard surgical procedure provided in the study by Chabaud-Williamson et al [13]. Moyambe JNT et al also preferred an adnexectomy in their case study [7]. Özcan R's team also described 03 cases of oophorectomy out of 12 due to the difficulties in finding a clear dissection plane between the mass and the ovary [6]. And in our case, we can ask ourselves this question: why had we not performed a cystectomy and preserved the ovary for better future fertility of this 13-year-old girl? In fact, it was impossible for us, intraoperatively, to identify healthy tissue in this tumor mass. Our action was further motivated by this rise in the CA125 level to approximately 5 times normal before the intervention.

Anatomypathology: An essential and indisputable examination, it represents the confirmatory diagnosis. In its pure form, mature cystic teratoma is benign; but although rare complication, malignant transformation can occur. It is also characterized by slow growth (1.8 mm per year) [10]. In the cases of Moyambe JNT et al; and Rousseau MC, the anatomical pathology concluded that it was a mature teratoma, although the latter revealed a very high level of CA 125 and a high level of CA 19.9 before the surgical intervention [3, 7]. We also had histological confirmation of mature ovarian teratoma in our case, with also a very high level of CA 125. As for the right ovary, histology revealed follicular cysts. What could then explain this discordance and especially this very high level of this marker in a confirmed mature teratoma? Rousseau MC also noted a fall or normalization of the markers one month after the intervention and explained this by a false increase due to peritoneal inflammation [14].

Follow up: Although the prognosis of mature ovarian teratomas is good or even excellent, their postoperative monitoring is necessary because cases of degeneration or recurrence or even new appearance have been reported by authors. Malignant transformation is rare, but it can occur in certain cases, particularly in women of advanced age [15]. Łuczak J's team noted metachronous disease of the contralateral ovary in two patients between 10 months and 05 years of postoperative follow-up [4]. She also reported normal menstruation in those with at least one ovary and cases of dysmenorrhea or simple ovarian cysts in some between 01 and 12 years of follow-up. Terenziani et al revealed 05 cases of metachronous ovarian disease including 04 after mature teratoma and 01 after immature teratoma [9].

I Guedira and his team reported an observation of a squamous cell carcinoma developed in a mature cystic teratoma of the ovary in a 46-year-old patient [5]. Xinge F et al also reported, although rare, a case of squamous cell carcinoma of the right ovary in a young 31-year-old patient who had as ATCD a cystectomy for mature ovarian teratoma, 06 years previously, a twin pregnancy by IVF/embryo transfer after 02 miscarriages and an ectopic pregnancy [14]. On the other hand, we did not find any documentation evaluating the prognosis of fertility in postoperative follow-up and this could be explained by the young age studied. In our case, the clinical and paraclinical examinations still did not note any particularity, with a normalization of the CA 125 in the second post-operative year [15].

4. Conclusion

Clinical exploration in cases of ovarian teratoma is still not easy, especially in adolescents or young girls. However, advances in medical science, in particular imaging and anatomical pathology, have revolutionized the diagnosis and management of these ovarian tumors. Faced with a large tumor without the possibility of identifying healthy ovarian tissue, a very high CA 125, we performed an adnexectomy. Histology confirmed a mature teratoma and follow-up up to 2 years did not note any particularity.

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