Massive Mature Cystic Teratoma Presenting as Ovarian Cancer Grade IIIC in a Young Vietnamese Woman: A Case Report

Huu Trung Nguyen[1], Ngoc Bich Trinh[1], Ngoc Hai Tran[2] and Phuc Nhon Nguyen[2,3*]

[1] Department of Emergency, Tu Du Hospital, Ho Chi Minh City, Vietnam

[2] Clinical Research Center (CRC), Tu Du Hospital, Ho Chi Minh City, Vietnam

[3] Department of High-Risk Pregnancy, Tu Du Hospital, Ho Chi Minh City, Vietnam

Received: 21 January 2025

Accepted: 16 February 2025

***Corresponding author:** docternhon@gmail.com

DOI 10.5001/omj.2028.17

Abstract

Ovarian teratoma is a common type of ovarian tumor. However, the particular appearance of ovarian teratoma and the presence of infiltrated lesions masquerading as a malignant tumor are rarely documented. Herein, we report an uncommon case at our consultant hospital and highlight the literature. A young Vietnamese female patient was hospitalized for an increased abdominal circumference and intermittent abdominal pain. The patient was managed with emergency surgery due to the suspicion of ruptured ovarian cysts resulting in abdominal hemorrhage. During surgery, bilateral teratoma ovarian tumors were suspected, however, the malignant lesion could not be completely excluded. Therefore, a management including bilateral salpingo-oovorectomy and partial resection of infiltrated omentum was done with a multidisciplinary team. The histopathological examination confirmed a benign lesion. The patient was discharged uneventfully. In conclusion, ovarian teratoma with a strange form ought to be ruled out for malignancy in young female patient. Interdisciplinary management is strongly recommended for a favorable outcome. Surgical management as malignant ovarian tumor could be fully considered. Histopathology is required to confirm diagnosis and further management. More studies are needed to unveil this extremely rare condition.

Keywords: Ovarian teratoma, multidisciplinary management, surgery, ultrasound, Vietnam.

Introduction

Ovarian teratomas are the most common ovarian germ cell tumors and account for 20% of all adult ovarian tumors.[1] This type of ovarian tumor is rarely related to malignant transformation and multidisciplinary input is generally not warranted.[2] They are composed of various tissues derived from one or more germ cell layers. At least two of three germ cell layers (ectoderm, mesoderm, and endoderm) are found within this tumor.[3]

Histologically, ectodermal derivatives are frequently the most prominent, which include keratinizing epidermis, sebaceous, sweat glands, hair follicles, and neuroectodermal components. Mesodermal derivatives include muscle, bone, cartilage, fat, and, occasionally, teeth. Endodermal derivatives include the thyroid and salivary gland, as well as respiratory and gastrointestinal tissues.[4] There are three major subtypes of ovarian teratomas including mature, immature, and monodermal teratomas.[1] These types could be presented in any stage of life, even in children and adolescents.[5],[6],[7]

Notably, mature teratomas are variable in size between a few cm to 15 cm (average diameter of 7 cm).[1] A massive size of ovarian teratoma is rarely reported.[8] The symptomatic manifestations are from mild to severe. Initial assessment is based on clinical examination. Imaging tools such as ultrasound (US), computed tomography (CT) scan, and magnetic resonance imaging (MRI) play an important role in diagnosing and disguising ovarian tumors with other pelvic tumors.[1] Ovarian mature cystic teratomas (MCTs) have wide spectrum of radiological presentation.[9] On the US, the tumors are characterized by echogenic sebaceous material, calcification, and nonspecific appearance such as heterogeneous, partially solid lesions, usually with scattered calcifications.[10] Histology and immunohistochemistry help identify the type of ovarian tumor.[11],[12]

Accordingly, proper management of ovarian germ cell neoplasm includes laparotomy and laparoscopy depending on the size of the tumor mass as well as the suspicion of malignancy. Apart from the benign tumor, the risk of malignant transformation increases in postmenopausal women if the surgical management is delayed.[13],[14] Thus, early management should be rigorously considered in young female patients. Hereby, we describe an uncommon case of ovarian teratoma presenting with a big size, strange shape, and appearance of ovarian cancer grade IIIC by macroscopic observation. Through this report, the team would like to underscore the need for individualized treatment strategies and the importance of early intervention as well as increasing awareness of clinicians.

Case Report

A 19-year-old Vietnamese girl was admitted to our Emergency Department for increased abdominal volume and pelvic pain evolving for several days. Her medical history of diseases was unremarkable. The patient declined sexual intercourse. Her menstrual period occurred frequently every 2-3 months. The first menstrual started at 13 years old. At admission, her vital signs were noted with a pulse rate of 122 bmp, blood pressure of 99/60 mmHg, respiratory rate of 20 times/minute, and body temperature of 37 Celsius degree. Her weight was 57 kg, her height was 158 cm, and her body mass index was 22.8 kg/m^2. The physical and gynecological examination revealed a palpable and painful mass on both sides of the pelvic cavity. The mass was solid and extended to the hypogastric region measuring 20 cm. Her serum laboratory tests are shown in Table 1. No infectious signs were detected. Her renal and liver function were completely normal.

Table 1: Serum laboratory tests.

Laboratory tests	Reference range	At hospitalization	After surgery
WBC (10^9/L)	3.37-8.38	10.12	13.59
NEU	39.8-70.5	71.6	78.0
RBC (10^{12}/L)	3.69-5.46	3.98	2.64
Hb (g/L)	108-164	122	80.0
Hct (L/L)	0.35-0.51	0.349	0.226
PLT (10^9/L)	172-378	306	323
Coagulation profile	Normal	Normal	Normal
Glycemia (mmol/L)	4.11-5.89	5.27	-
Ure (mmol/L)	2.76-8.07	4.2	-
Creatinine (μmol/L)	44-80	63	-
eGFR (mL/min/1.73m^2)	>60	112.23	-
AST (U/L)	<35	13.81	-
ALT (U/L)	<35	8.11	-
Na+ (mmol/L)	136-146	-	139.4
Potassium(mmol/L)	3.4-4.5	-	3.23
Chloride (mmol/L)	101-109	-	104.4
Calcium (mmol/L)	2.2-2.65	-	2.10
CA125 (UI/ml)	<35	52.3	-
HE4 (pmol/l)	<70	43	-
ROMA value (%)	<25.3	5.73	-
AFP (ng/ml)	0.89-8.78	<2.00	-

Beta HCG (mUI/mL)	<5	Negative	-

ALT: alanine transaminase; AFP: alpha-fetoprotein; AST: aspartate transferase; CA 125: carbo-hydrate antigen 125; eGFR: estimated glomerular filtration rate; Hb: Hemoglobin; HCG: human chorionic gonadotropin; Hct: hematocrit; HE4: Human epididymal protein 4; PLT: platelet; NEU: neutrophile; RBC: red blood cell; ROMA: risk of ovarian malignancy algorithm; WBC: white blood cell.

Ultrasound findings revealed a vide uterine cavity and a heterogenous mass measuring 195 x 60 x 160 mm without a solid component and hypervascularity. Inside the tumor, shadowing echogenicity was observed. The borderline of the ovarian tumor was ill-defined. The intraabdominal fluid collection was detected at a large volume. A diagnosis of ruptured ovarian teratoma was made (Fig 1). The malignant bilan of the ovarian tumor was seemingly negative. Immediately, the patient was transferred to the operating room for an emergency exploratory laparotomy decision.

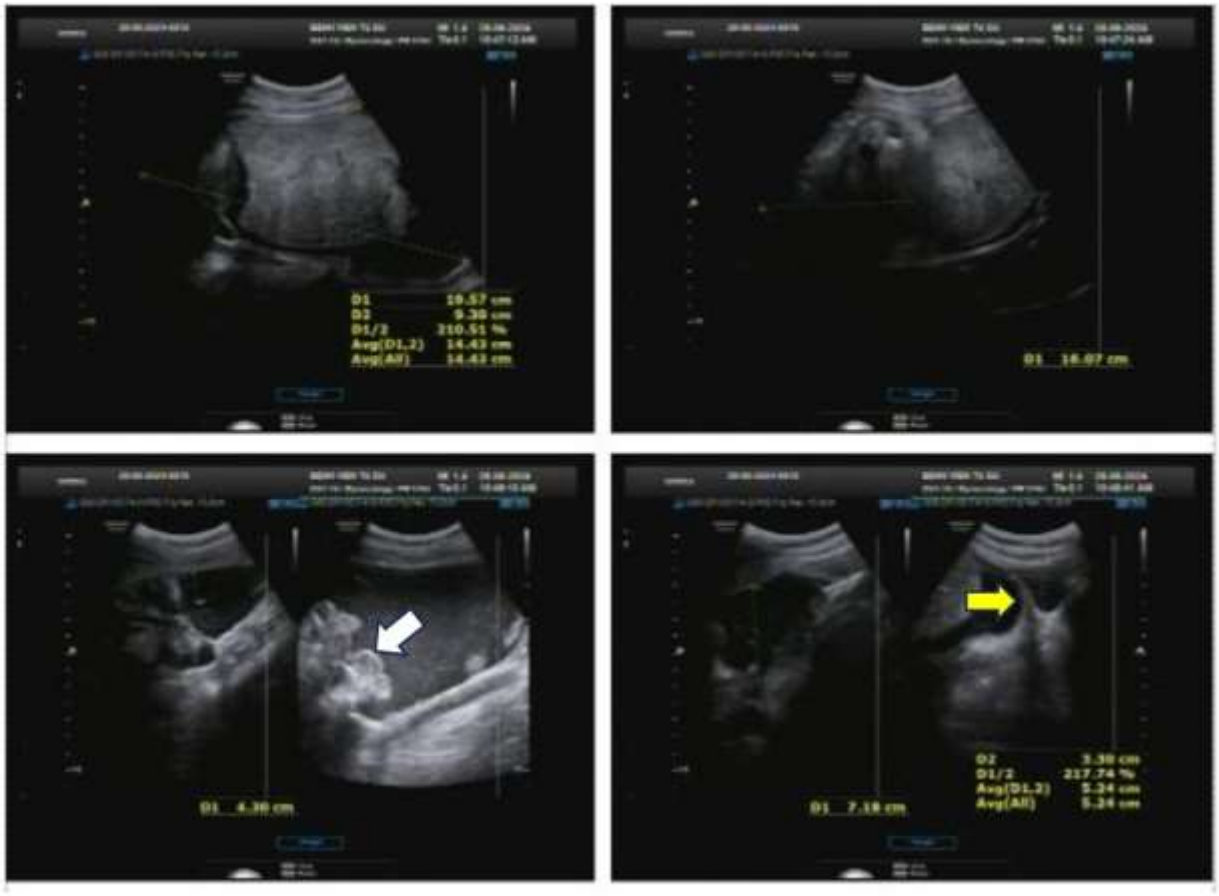

Figure 1: Ultrasound shows mural hyperechoic Rokitansky nodule or dermoid plug (white arrow), shadowing calcific or dental (tooth) components, the presence of fluid-fluid levels, and septal structure (yellow arrow). No internal vascularity was found on the color Doppler signal.

After counseling, a sub-through-umbilical incision laparotomy was performed. Upon laparotomy, the abdominal cavity contained the diffused-yellow fluid and adherent structures. After releasing adhesion, the uterus was normal, the bilateral annexes were occupied by massive tumors measuring 20x25 cm in size. This large ovoid- to bilobed-shaped cystic pelvic structure extends to the lower abdomen. Dermoid cysts were detected with the intracystic component of teeth, hair, skin, and mixtures of fat and water (Fig 2 and 3). Seriously, the fallopian tubes were adherent severely to the ovarian tumor on the left-side and the right-side ovarian tube was dilated. Nodule-infiltrated lesions were found at the abdominal wall and the omentum.

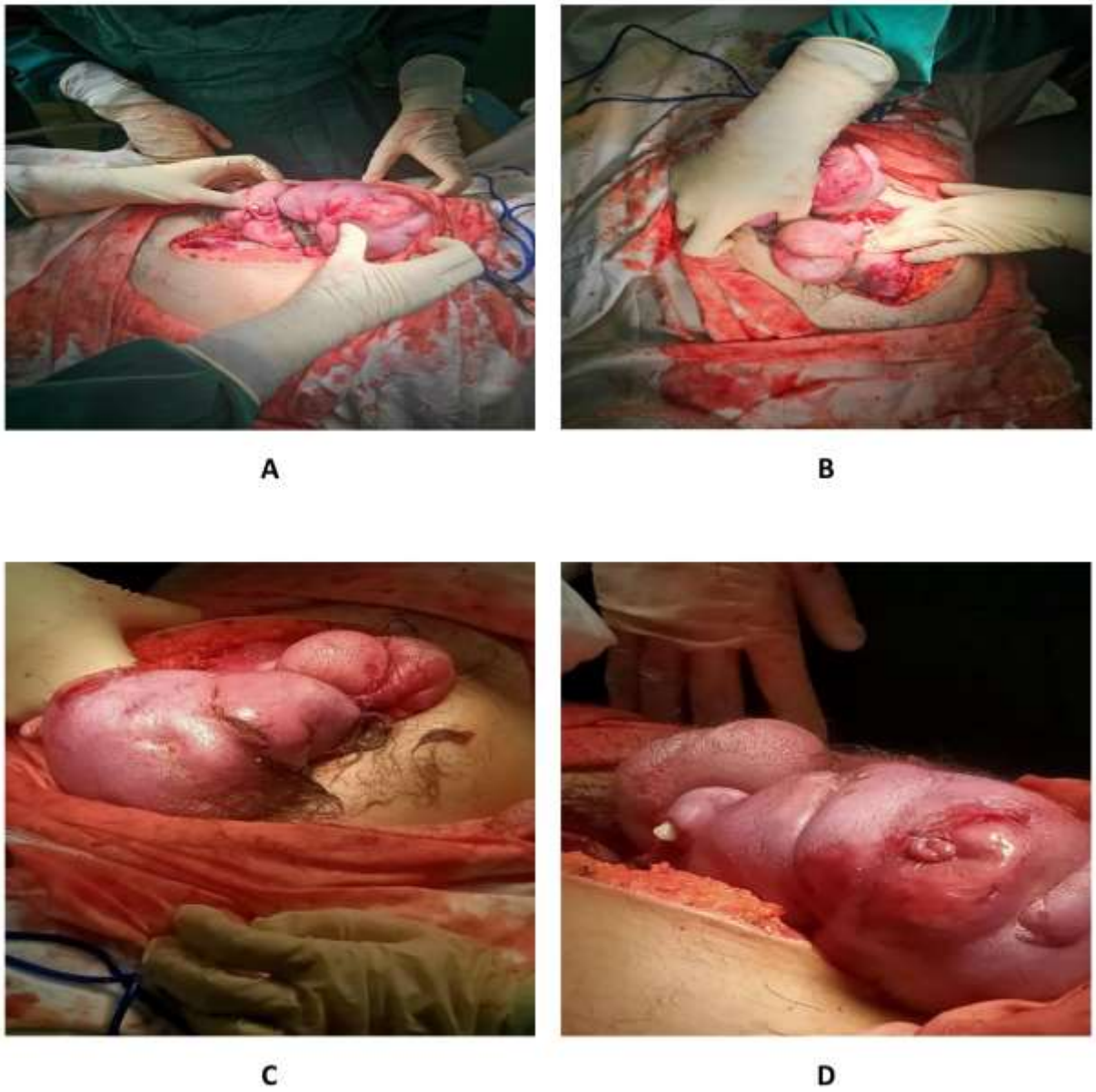

Figure 2: Intraoperative photos show strange tumors arising from both ovaries (A: the right side, B: the left side). The tumor is covered by skin, presenting with hair and teeth-like structure (C-D).

Figure 3: The macroscopic specimen shows a yellowish fat component, cartilage, cheese-like material (creamy keratin), and fine hair shafts arising from the nodule.

Therefore, the digestive surgeon was invited to join the surgery. Firstly, ovarian-sparing surgery was done. However, due to the adherent complication and the atypical presentation of the ovarian tumor, bilateral salpingo-oovorectomy were performed. A part of the infiltrated omentum was relevant for frozen section biopsy. The total blood loss was 400 ml. An abdominal drainage was placed. The chemical peritonitis was prevented with antibiotic prophylaxis. Nevertheless, the postoperative course was marked with subcutaneous infection. However, the infection was responded well to the spectrum-broad antibiotic therapy in 5 days. The Douglas pouch and abdominal drainage were removed on postoperative day 3. The patient was discharged uneventfully. Histopathological results confirmed benign ovarian tubes, teratoma ovarian tumor, and inflammatory reaction of the omentum (Fig 4 and 5). The patient was followed up for 2 months without surgical complications. The patient and her mother were thankful to our team for the treatment.

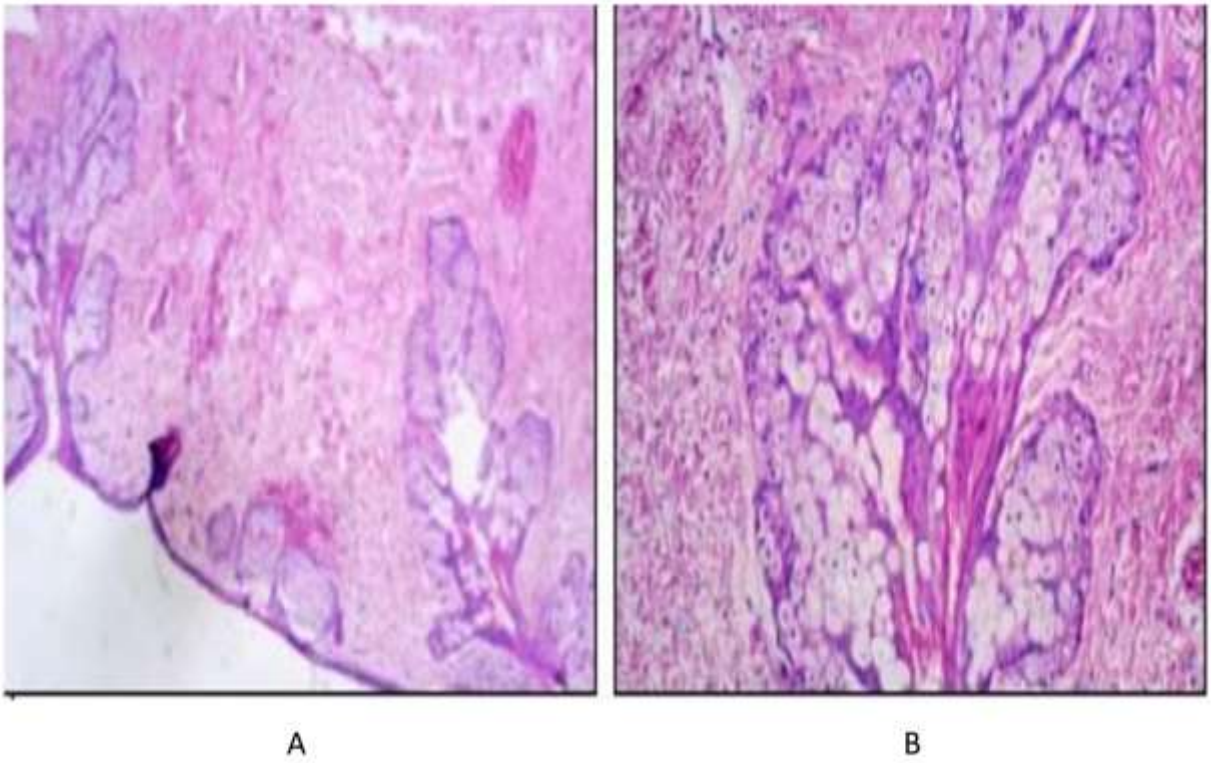

Figure 4: Photomicrograph of mature teratoma shows the presence of skin, sebaceous gland, and hair follicles (H and E, original magnification ×10 (A), x40 (B), respectively).

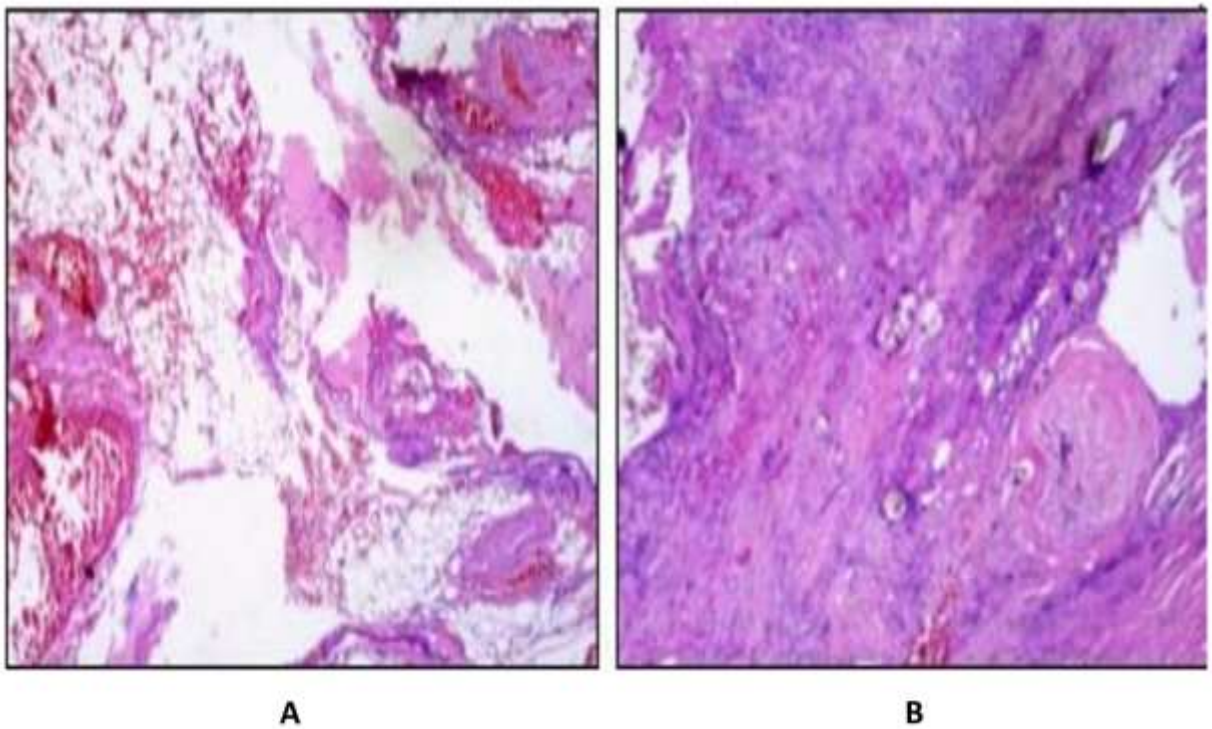

Figure 5: Photomicrograph of the omentum contains the presence of hair-like structure and inflammatory tissue. (H and E, original magnification ×10 (A), x40 (B), respectively).

Discussion

Ovarian teratomas include mature cystic teratomas (dermoid cysts), immature teratomas, and monodermal teratomas (struma ovarii, carcinoid tumors, neural tumors).[10] The symptoms of ovarian teratomas are present in various manifestations including asymptomatic presentation and severe complications such as malignant transformation, torsion, rupture, and hemorrhage.[15] Shadowing echogenicity named Rokitansky nodule or dermoid plug is described as a densely echogenic tubercule projecting into the cystic lumen. This sign is frequent in 81-86% of ovarian teratomas.[9] Although the benign images on ultrasound and biochemical markers were found, the diagnosis of benign and malignant ovarian tumor in our case is completely crucial due to the malignant appearance of the ovarian tumors at laparotomy. Imaging features of immature and monodermal teratomas are less specific, but a combination of clinical features and imaging findings can help in the diagnosis.[1] As mentioned above, the initial ultrasound suspected a teratoma ovarian tumor. Since the ruptured complication was diagnosed, the emergent surgery was performed without further investigation by magnetic resonance imaging or computed tomography scan of the chest, abdomen, and pelvis before surgery.

Due to the scarcity of data, current understanding of each subtype is limited and treatment has generally been derived from the more common tumor types.[12] Immature ovarian teratomas are a rare subtype of germ cell tumors characterized by the presence of embryonic elements, particularly primitive neuroepithelium, and they typically affect young women.[16] Especially, a well-differentiated cerebellum within a mature cystic teratoma of the ovary has been reported.[17],[18] Recently, AlEssa et al., have documented a case of well-differentiated cerebellum within ovarian teratoma which had a similar appearance with our case.[19] Anti-N-methyl-D-aspartate receptor (NMDAR) encephalitis relating to ovarian tumor has been also documented at our tertiary referral hospital.[20]

Laparoscopic surgery could be applied in almost all cases of ovarian tumors with a lower surgical complication rate.[15] However, this surgical method could not be performed well with manipulation for huge-size tumors. Moreover, this advanced technique should be rigorously in the suspicion of ovarian cancer. Rarely, a malignant transformation of ovarian mature cystic teratoma (MCT) has occurred at trocar scar in a young woman following laparoscopic ovarian cystectomy.[21] In general, ovarian-sparing surgery should be evaluated and performed carefully in young patient for future fertility, avoiding adnexectomy. However, a strict monitoring after surgical intervention should be established. According to Luczak et al., predominantly solid structures noted on imaging studies, large dimensions, and positive tumor markers are clinical predictors of malignancy. A diagnosis of purely cystic lesions with negative markers or of a small size should be an indication of a gonad-sparing procedure.[22] Understanding uncommon findings as well as classic signs with basic knowledge of pathological equivalents permits a more accurate diagnosis and guides adequate treatment.[9] Importantly, gross examination along with adequate sampling of all solid and suspicious areas in the ovarian cyst wall are necessary to avoid missing any rare or synchronous malignancies.[23]

Conclusion

Malignancy should be ruled out from ovarian teratoma with atypical presentation in a young female patient. Raised awareness of clinicians is completely necessary. A histopathological examination is required for the identified diagnosis. Interdisciplinary management is strongly recommended for a favorable outcome. A surgical management similar to the treatment of malignant ovarian tumors ought to be fully considered. Due to the paucity of data, this presence of ovarian tumors is not well documented. Further data is needed to better understand this extremely rare entity.

References

1. Saleh M, Bhosale P, Menias CO, Ramalingam P, Jensen C, Iyer R, et al. Ovarian teratomas: clinical features, imaging findings and management. Abdom Radiol (NY) 2021 Jun;46(6):2293-2307.

2. Ozgur T, Atik E, Silfeler DB, Toprak S. Mature cystic teratomas in our series with review of the literature and retrospective analysis. Arch Gynecol Obstet 2012 Apr;285(4):1099-1101.

3. Srisajjakul S, Prapaisilp P, Bangchokdee S. Imaging features of unusual lesions and complications associated with ovarian mature cystic teratoma. Clin Imaging 2019;57:115-123.

4. Desouki MM, Fadare O, Chamberlain BK, Shakir N, Kanbour-Shakir A. Malignancy associated with ovarian teratomas: frequency, histotypes, and diagnostic accuracy of intraoperative consultation. Ann Diagn Pathol 2015 Jun;19(3):103-106.

5. Mremi A, Rwenyagila D, Chilonga K, Sadiq A, Msuya D, Lodhia J. Immature teratoma of the ovary in a 1 year and 9-month-old child: a case report and review of the literature. J Surg Case Rep 2021 Jan;2021(1):rjaa609.

6. Gkrozou F, Tsonis O, Vatopoulou A, Galaziou G, Paschopoulos M. Ovarian Teratomas in Children and Adolescents: Our Own Experience and Review of Literature. Children (Basel) 2022 Oct;9(10):1571.

7. Fiegel HC, Gfroerer S, Theilen TM, Friedmacher F, Rolle U. Ovarian lesions and tumors in infants and older children. Innov Surg Sci 2021 Aug;6(4):173-179.

8. Tao J, Shi Z, Li M, Li T. Growing teratoma syndrome of the ovary: a case report and literature review. Front Oncol 2024 Jul;14:1412206.

9. Sahin H, Abdullazade S, Sanci M. Mature cystic teratoma of the ovary: a cutting edge overview on imaging features. Insights Imaging 2017 Apr;8(2):227-241.

10. Outwater EK, Siegelman ES, Hunt JL. Ovarian teratomas: tumor types and imaging characteristics. Radiographics 2001;21(2):475-490.

11. Cong L, Wang S, Yeung SY, Lee JH, Chung JP, Chan DY. Mature Cystic Teratoma: An Integrated Review. Int J Mol Sci 2023 Mar;24(7):6141.

12. Debuquoy C, Romeo C, Vanacker H, Ray-Coquard I. Rare ovarian tumors: an update on diagnosis and treatment. Int J Gynecol Cancer 2020 Jun;30(6):879-887.

13. Mahtate M, Talib S, Slaoui A, Zeraidi N, Lakhdar A, Rhrab B, et al. Malignant Degeneration of a Mature Ovarian Teratoma. Case Rep Obstet Gynecol 2021 Jul;2021:5527467.

14. Rathore R, Sharma S, Agarwal S. Malignant transformation in mature cystic teratoma of the ovary: a retrospective study of eight cases and review of literature. Przeglad menopauzalny = Menopause review. 2018;17(2):63-8.

15. Tankou Ja, Foley OW, Liu CY, Melamed A, Schantz-Dunn J. Dermoid cyst management and outcomes: a review of over 1000 cases at a single institution. American Journal of Obstetrics & Gynecology. 2024;231(4):442.e1-.e7.

16. Errami L, Douzi A, Lamzouri O, Taheri H, Saadi H, Mimouni A. Immature ovarian teratoma in a 20-year-old woman: A case report. J Int Med Res 2024 Feb;52(2):3000605241232568.

17. Remadi S, Burkhardt K, Straccia AT, Pizzolato G, Mac Gee W. Well differentiated cerebellar tissue within a mature cystic teratoma. Pathol Res Pract 1998;194(5):371-374.

18. Shintaku M, Sakuma T, Ohbayashi C, Maruo M. Well-formed cerebellum and brainstem-like structures in a mature ovarian teratoma: Neuropathological observations. Neuropathology 2017 Apr;37(2):122-128.

19. AlEssa A, AlAhmadi HH, Ahmed A, AlMousa A, Hamadeh NG, Farah YA. Well-differentiated cerebellum in an ovarian mature cystic teratoma: a case report and review of the literature. J Med Case Rep 2022 Jun;16(1):215.

20. Pham TH, Nguyen PN, Ho QN. Timely Laparoscopic Intervention for Ovarian Tumor-related Autoimmune Encephalitis: A Challenging Pathology at Tu Du Hospital in Vietnam and Literature Review. Gynecol Minim Invasive Ther 2023 Aug;12(3):185-188.

21. Okuda T, Uda Y, Sakai S, Harada T. Malignant Transformation of Unknown Duration of an Ovarian Mature Cystic Teratoma Presenting as a Trocar Recurrence in a Young Patient: A Case Report and Literature Review. Case Rep Obstet Gynecol 2023 Nov;2023:8875092.

22. Łuczak J, Bagłaj M. Selecting treatment method for ovarian masses in children - 24 years of experience. J Ovarian Res 2017 Sep;10(1):59.

23. Al Harthi S, Al Kalabni M, Al Qassabi B, Al Ajmi R. Mucinous Adenocarcinoma and Carcinoid Tumor Arising Within an Ovarian Mature Cystic Teratoma: A Case Report. Oman Med J 2023 Jul;38(4):e538.