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REVIEW ARTICLE

Female Adnexal Tumor of Probable Wolffian Origin (FATWO): Case Report and Systematic Literature Review

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Abstract

Purpose The paper aims to provide additional evidence and summarize the available evidence concerning the diagnosis, treatment, and prognosis of female adnexal tumors of probable Wolffian origin (FATWO).

Methods We presented a case of FATWO diagnosed and treated at our department. In addition, we performed systematic research of the literature (Scopus, PubMed/MEDLINE, ScienceDirect, and the Cochrane Library) including observational prospective and retrospective studies, case series, and case reports. We collected 165 cases of FATWO are available in the literature, including our case report.

Results We presented a case report of a 52-year-old asymptomatic woman with an incidental finding of an ovarian cyst. The final histological examination diagnosed it as an ovarian FATWO. Following surgical treatment alone, the patient continued to have negative follow-ups. The median age at diagnosis was 50 years old; the most frequent localization was the broad ligament (30,3%) followed by the ovary (26.1%), mesosalpinx (21.8%), and less common localizations. The most frequent symptom at the diagnosis was abdominal pain. Recurrent disease occurs in less than 20% of the cases with an overall survival of 59 months in patients with recurrent disease.

Conclusions FATWO is a rare cancer arising from mesonephric remnant most frequently from the broad ligament.

Keywords Ovarian tumor · Female adnexal tumor of probable Wolffian origin · FATWO · Rare cancer

Introduction

Female adnexal tumors of probable Wolffian origin (FATWO) are rare gynecological tumors, and the first case was described by Kariminejad and Scully in 1973 [1, 2]. During female embryonic development, a regression of the mesonephric or Wolffian ducts occurs but remnants of these structures could be found postnatally in the uterus, vagina, adnexa, and broad ligament [3, 4]. It was postulated that FATWO derived from the aforementioned embryonic structures. Different terms were used in the literature to describe them, and recently, the World Health Organization introduced in its classification of female tumors the term Wolffian tumor [5, 6]. More frequently, FATWO arose from the broad ligament but origin from the ovary, tube, mesosalpinx peritoneum, or paravaginal tissue was reported in the literature [7]. These neoplasms are globally considered as low malignant potential, and the majority of the cases had a benign behavior [2]. However, malignant behavior or recurrences of disease were described. We reported a case of ovary FATWO managed in our institution. Moreover, to elucidate the clinicopathologic and immunohistochemical features of a such rare tumor, we performed a systematic review of the literature.

Methods

We conducted a systematic review of the literature performing a literature search in the electronic databases Scopus, PubMed/MEDLINE, and ScienceDirect from the database

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inception to November 2023. The search strategy included the combinations of the medical terms "Ovarian tumor," "female adnexal tumor of Wolffian origin," and "tumor of Wolffian origin." We included in the systematic review all papers published in the English language reporting at least one case of uterine FATWO confirmed at the pathological evaluation. Given the rarity of the topic, the authors included case reports and case series. This review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and was registered in the International Prospective Register of Systematic Reviews (PROSPERO, ID CRD42024573665).

Study Selection

Titles and/or abstracts of identified studies were screened independently by two authors (FFA and FC). The full text of the potentially eligible studies was retrieved and independently assessed for eligibility by other two review team members (FF and SG). Any disagreement over the eligibility of studies was resolved through discussion with a fourth author (FO). The references of all identified studies were systematically revised to identify other eligible publications. Moreover, identified literature reviews were retrieved to search for possible additional publications in the reference lists. The review was written following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [8].

Data Extraction

A standardized form was used to extract data from included studies. For each reported case, we collected demographic characteristics, presentation symptoms, surgical data, immunohistochemical and pathological features, treatment strategy, and follow-up details. Standard descriptive statistics were used to summarize the characteristics of identified cases as appropriate.

Case Report

In September 2021, a 52-year-old G1, P1 postmenopausal woman (since 2018), with no prior surgical history or family history of malignancy, was referred to our center following the detection of a right ovarian cyst during a routine gynecological ultrasound screening. At the time of the diagnosis, the tumor markers were normal (AFP 4.1 U/mL, CA15.3 22.6 U/mL, CA19.9 1.8 U/mL, CA125 9.7 U/mL, and HE4 37.6 U/mL), and the patient was asymptomatic. A color Doppler ultrasonography was performed by an expert operator, and the right ovarian mass was characterized by a multilocular-solid cyst, with a diameter of 34 × 34 × 32 mm. Doppler examination revealed minimal blood flow (color score 2). The uterus and the left ovary were normal, and there was no free fluid (Fig. 1).

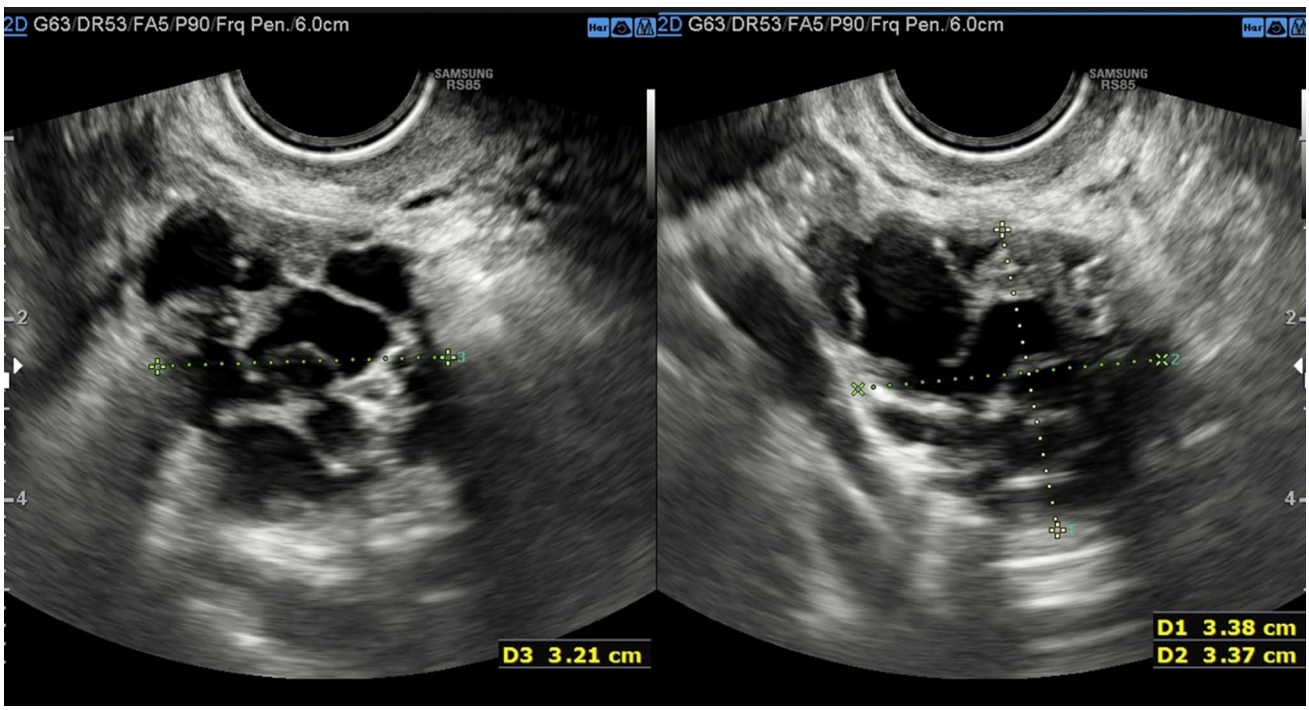

Fig. 1 Preoperative ultrasound image of ovarian mass

In October 2021, the patient underwent a planned laparoscopic bilateral salpingo-oophorectomy, peritoneal washing, and concurrent endometrial biopsy. During the surgery, the ovarian mass was confirmed, and no other localizations were observed. Histological examination unveiled a neoplasm with a combination of solid, trabecular, and tubule-cystic growth patterns, the latter less represented with eosinophilic, colloid-like material in most of the tubule-cystic lumens. Stroma was scant in all the growth patterns with a focal myxoid appearance. Neoplastic cells showed mainly a round to polygonal morphology, with pale to eosinophilic cytoplasm and a bland, centrally located nucleus with some small nucleoli visible. A flat morphology was detected within the luminal layer whereas a spindle cell component was present in the solid areas. Very rare mitoses were detected. Immunohistochemistry showed strong and diffuse positivity for calretinin, CD10, and WT1, whereas a patchy staining pattern for cytokeratin AE1/AE3 and inhibin was detected; vimentin showed a diffuse positivity but with very variable intensity; EMA was negative (Fig. 2).

The final histopathological diagnosis was a female adnexal tumor of probable Wolffian origin (FATWOs) on the right ovary and a placental side nodule on the endometrial biopsy. The left adnexa and the cytology were negative. After a multidisciplinary discussion with the gynecological, oncological, and radiotherapist group, a second surgery was planned. Therefore, in January 2022, a total hysterectomy, multiple omental and peritoneal biopsies (vesicouterine

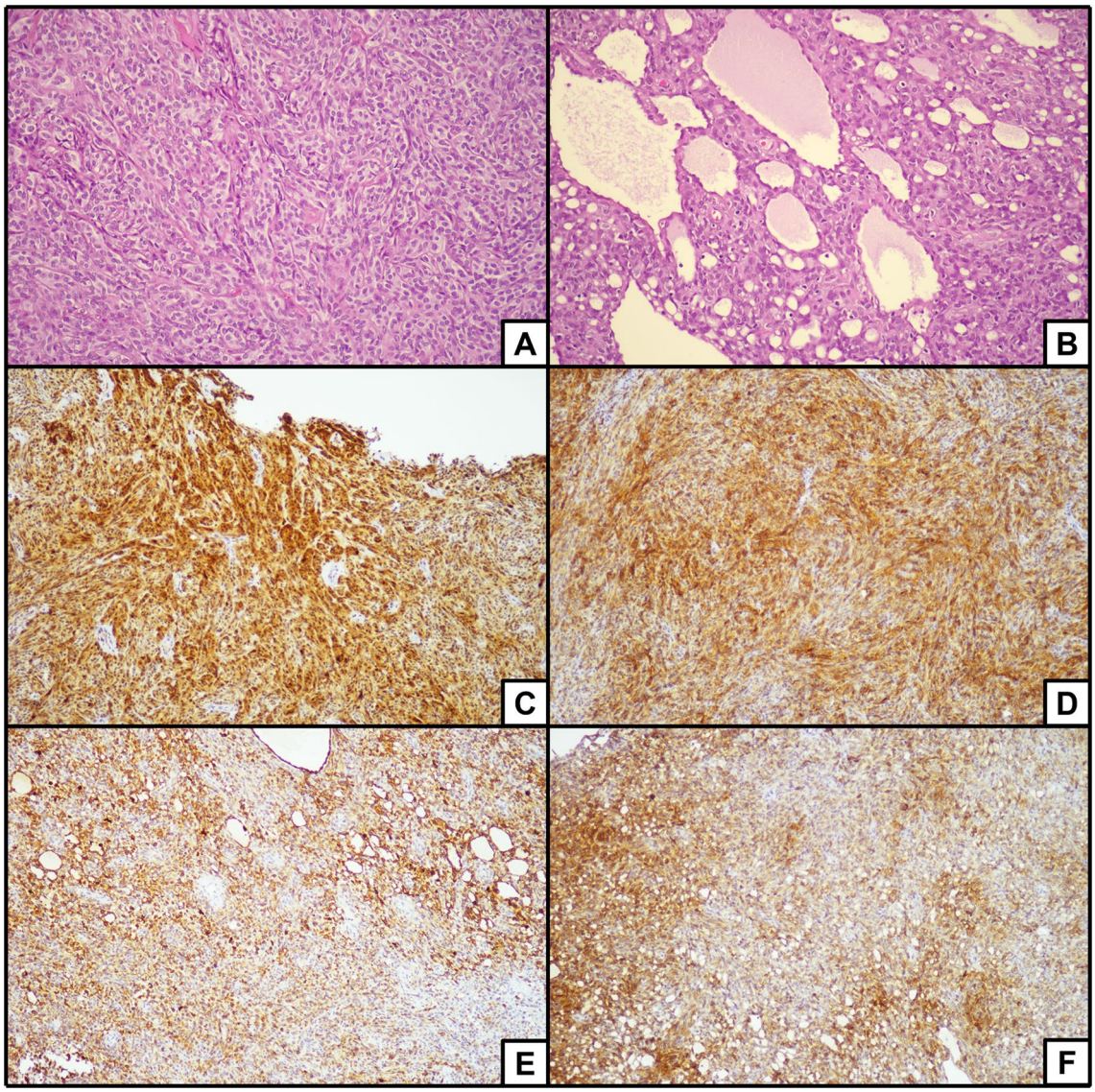

Fig. 2 H&E stained sections showing solid cords growth pattern (**a**) and tubulocystic growth pattern (**b**). 20x. Immunohistochemistry showing expression of calretinin (**c**), CD10 (**d**), cytokeratin AE1/AE3 (**e**), and inhibin (**f**). 10x

fold, Douglas cord, and bilateral paracolic gutter), and peritoneal washing were performed. At the definitive uterine histopathological analysis, the surgical specimen, biopsies, and the cytology were negative. The postoperative course was regular without any complications, and the patient was dismissed on the 2nd postoperative day. After a second multidisciplinary discussion, the treatment was considered concluded with surgery, and the patient was advised to follow up every 6 months. At the time of publication, the patient is alive without any localization of disease.

Results

Literature Search Results

The search strategy in the abovementioned electronic bibliographic databases retrieved 243 items. After removing duplicates (n = 56), out of 187 potentially eligible studies, 113 studies were excluded because not reporting cases of FATWO. Finally, we included 74 studies in our review, for a total of 165 cases of FATWO. The flowchart of study selection is shown in Fig. 3. The results of the literature review are summarized in Table S1.

Clinical Features

The age of the patients ranged from 13 to 83 years old, and a median age of 50 years (IQR 38–81) was reported. The clinical presentation was heterogeneous and unspecific and in 51 cases was not reported. In 42,6% of the patients were asymptomatic, and FATWO was an incidental finding. The most common symptoms were pelvic or abdominal pain (45.2%), vaginal bleeding (7.8%), sporadically reported ascites, urinary retention, tenesmus, and pleural effusion. The mean size was 87 mm, and the broad ligament was the most frequent localization (30,3%) followed by the ovary (26.1%), mesosalpinx (21.8%), paratubal site (6.7%), tubes (5.5%), unspecified adnexal localization (3.6%), and others rare sites (6.0%) as Douglas, vaginal apex, retroperitoneum, pelvic wall, and paravaginal space. In 12 cases (7.3%), metastasis was reported at the time of initial diagnosis: omentum in five cases, peritoneum implants in six cases, and peri-hepatic and parenchymal lung in one case, respectively. Specific markers were not identified in the literature review, and CA-125 was elevated only in the 3% of the FATWO.

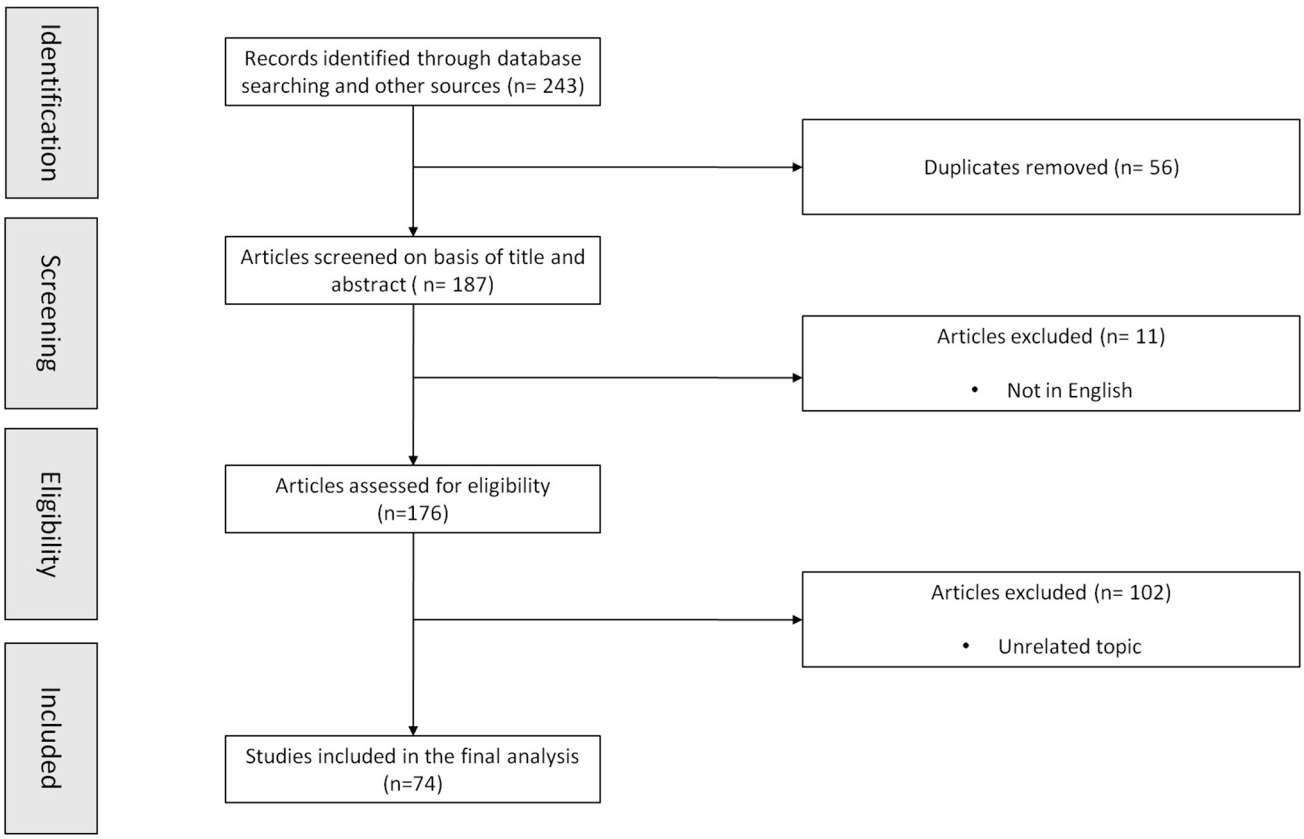

Fig. 3 Flowchart of studies selection

Treatment Modalities

The review of the literature did not identify a standardized treatment approach and in 39.7% of patients was not reported. In the other cases, first-line treatment was surgery, mostly with a laparotomic approach (83%). In the reported cases, the proposed treatment was hysterectomy with bilateral adnexectomy (46%), bilateral salpingo-oophorectomy (11%), monolateral salpingo-oophorectomy (14%), resection of the mass (18%), salpingectomy or partial salpingectomy (3%), and anterior exenteration (1%). Additional surgical procedures were reported: omentectomy (20%), pelvic (8%) and para-aortic (5%) lymphadenectomy, resection of peritoneal implants (4%), appendectomy, partial colectomy, and sigmoid resection. After primary surgery, adjuvant platinum-based chemotherapy was administered in five cases (5%), radiotherapy and tamoxifen in two and one patient, respectively.

Follow-Up and Oncological Outcomes

Follow-up data were reported in 57 patients, and median length was 38.5 months (IQR 12–60). Recurrence of disease was reported in 29 cases (17.5%), three of which presented an extra-adnexal localization at the time of the first diagnosis. Median progression-free survival (PFS) at the first recurrence was 22.5 months (IQR 9.75–36.3), and overall survival in patients with recurrent disease was 59 months. Recurrent disease developed in patients who initially received only tumor resection (25%). The most frequent sites of recurrence were the peritoneum (31.0%), liver (20.7%), broad ligament (17.2%), omentum (10.3%), and other rare localization as an umbilical port, paravaginal space, vaginal cuff, and bowel mesentery. Surgery was performed in 20 cases (69%) followed by medical or chemoradiation therapy in five and two cases, respectively. Treatment of metastatic disease involved the use of tyrosine kinase inhibitor (t-Kit) in four patients.

Immunochemistry

Immunohistochemical staining was not reported in 49 cases. The results of the literature review are summarized in Table 1.

Table 1 Immuhistochemical profile of FATWO cases

Markers	Positive/tested	Positive rate (%)
Calretinin	73/90	81.0
Vimentin	73/97	75.2
Inhibin	51/86	59.3
Broad-spectrum cytokeratin	74/81	91.0
Cytokeratin 7	52/66	78.8
CD 10	40/58	69.0
Estrogen	27/78	34.6
Progesterone	21/53	39.5
WT 1	23/55	41.8
PAX 8	4/17	23.5

Discussion

Our systematic literature review included 166 cases of FATWO reporting a substantially higher number of cases compared to other recent reviews [9–12]. Wolffian neoplasms were often diagnosed during gynecological routine visits in asymptomatic patients. The mean size was 87 mm and could partially explain the pelvic or abdominal pain reported in up to 45% of the cases. The mass is typically unilateral with only two cases of bilateral tumors in which metastatic spreading to the controlateral side could not be excluded [13, 14]. The age of patients diagnosed with FATWO ranged from 13 to 83 years old but women older than 50 years were likely to develop this rare tumor.

FATWO diagnosis is challenging due to the common features of other gynecological entities and the absence of a well-defined immunohistochemical profile (IHC) [12, 15]. Certainly, the broad ligament localization of a mass could be suspicious for FATWO but, as reported in our case report, ovarian and mesosalpinx localization are less uncommon than reported by other authors [9, 13]. The final diagnosis of FATWO included the findings of tubular and glandular structures and the presence of eosinophilic amorphous secretions in the glandular lumen [9]. Based on the results of the literature review, a specific immunohistochemical panel was not identified but IHC is fundamental for the diagnosis. We reported an immunopositivity for calretinin (81%), vimentin (75.2%), inhibin (59.3%), broad-spectrum cytokeratin (91%), CK7 (78.8%), CD10 (69%), and WT1 (41.8%). Usually, FATWO was negative for actin, EMA, S100, and human bone marrow endothelial marker-1 (HBME-1). We reported a lower positivity for estrogen (34.6%) and progesterone (39.5%) compared to Bennet et al. case series [15]. Mesonephric remnants are usually PAX8 negative but we reported four cases of positivity [2, 15, 16].

Differential diagnosis covers a large spectrum of gynecological tumors including endometrioid carcinoma, Sertoli–Leyding cell tumors (SLCT), granulose cell tumors (GCT), and metastatic cancer from non-gynecologic organs. In consideration of the variety of overlapping histological features, the role of intraoperative pathologic examinations is limited. Endometrioid carcinoma arises in a background of endometriosis, present cellular atypia, nuclear pleomorphism, and squamous/mucinous differentiation [15, 17]. SLCTs present Sertoli–Leydig cells, negativity for

inhibin, calretinin, WT-1, CD10, and positivity for epithelial membrane antigen and are often characterized by endocrine symptoms [15, 18, 19]. GCTs determine endocrine symptomatology, anatomopathological examination shows grooved nuclei and sparse cytoplasm of tumor cells, and immunochemistry is normally negative for CK 7 [7, 20].

In the majority of the cases, an abdominal hysterectomy plus bilateral adnexectomy, or debulking of the tumor if necessary, was performed, and it was considered the most appropriate treatment for primary FATWO, according also to our management. No case of nodal recurrence was reported, and we do not suggest systematic pelvic or paraortic lymphadenectomy. Otherwise, omentectomy was performed in 20% of the cases, and despite the omentum being a possible site of secondary implants, the resection without macroscopic localization is controversial. Although some authors reported adjuvant therapy after primary surgical resection (8%) chemo and radiation therapy was not widely administered, and it was not routinely considered.

The Wolffian neoplasms are considered to have a low malignant potential and to present an indolent behavior. We found a 17% risk of recurrence with 29 cases reported in the literature, higher than Shalaby et al. [12] results and following Chen et al. [2]. The median PFS was 24 months, and it was shorter than the results of the partial analysis of FATWO cases provided by other authors [4, 21, 22]. Identifying the malignant cases may lead to more aggressive surgical management, consideration of adjuvant chemotherapy, closer follow-up and adequate counseling. Conversely to other rare gynecological tumors, validated anatomopathological scores were not available, and the distinction between benign and malignant is mainly related to the clinical behavior [23]. Some authors proposed criteria of malignancy including tumor size greater than 10 cm, capsular invasion, capsular rupture, a high number of mitoses, cellular pleomorphism, and macroscopic tumor implants [12, 21, 24, 25] but recurrence seemed to occur also in patients without aggressive histopathologic findings [12, 26]. Nakamura et al. suggested a possible diagnostic role of CD 56 as a marker of malignancy but our review could not confirm the hypothesis [27]. In 25% of the recurrence diseases, a simple tumor resection was initially performed, and we can speculate that a conservative surgical approach could represent a risk factor for relapse and needs to be taken into account when fertility-sparing treatment is proposed. Based on our results, five cases of disease-related death were reported with a median overall survival of 36 months (IQR 27–48); nonetheless, the findings could suffer from a lack of follow-up data. In 71.4% of the recurrent diseases, surgical resection was performed followed by adjuvant medical therapy or chemoradiation therapy in 65% and 10% of patients, respectively. Chemotherapy and radiation therapy demonstrated a controversial role in the treatment of recurrences, and different regimens were used. The most frequent used first-line regimens were carboplatin/paclitaxel [2, 7, 9, 26, 28–30], cisplatin/cyclophosphamide [26, 31], cisplatin/doxorubicin [32], and cisplatin/oxaliplatin/docetaxel; as second-line treatment was administered topoisomerase I inhibitor [30, 31, 33], carboplatinum/etoposide [30], gemcitabine [33], and 5-fluorouracil. The association of carboplatin/paclitaxel demonstrated a stabilization of the disease in patients with metastatic disease but the heterogeneity in doses and regimens did not allow for conclusions [2, 26]. Wolffian tumors with a c-kit positive profile may benefit from treatment with c-kit tyrosine kinase inhibitors, and different authors reported a good response in metastatic disease [29, 31, 34, 35]. STK11 mutation was identified in some cases of FATWO and testing with next-generation sequencing could permit to selection of candidate patients to c-kit inhibitors [15, 34].

In conclusion, FATWO are rare gynecological cancers, the clinical presentation is unspecific, and patients are often asymptomatic. The majority of Wolffian tumors are benign but a 17% recurrence rate has been reported. Diagnosis included anatomopathological and immunohistochemical features that allow it to differentiate from other tumors. Although there is no consensus on the treatment, surgical resection, preferably hysterectomy, and bilateral adnexectomy, if not desire of fertility-sparing, is the first choice. All future cases should be reported and published, and mutational analysis is advised to better categorize FATWO genetic profile and evaluate the role of different target therapies.

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Author's Contribution All the authors conform to the International Committee of Medical Journal Editors (ICMJE) criteria for authorship, contributed to the intellectual content of the study, and approved the final version of the article. FFA and FC conceptualized and designed the study. FC and GE revised medical records and collected the data. FFA, FC, and LA performed a systematic review of the literature and extracted the data. FFA, FF, and SG managed the dataset and performed statistical analyses. FFA, FF, SU, SG, MF, ES, and FO wrote the manuscript. All authors contributed to the interpretation of the results and the writing and editing of the manuscript.

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Declarations

Conflict of interest None. The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Ethical Standards The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Hel-

sinki Declaration of 1975, as revised in 2008. All included participants gave consent for case presentation and anonymized data collection and analysis for research purposes.

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