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Case Report

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Ovarian Gynandroblastoma Presenting with Pleural Effusion: A Case Report and Literature Review

Anna Rita Palumbo*, Valentino De Vivo, Fiorina Citarella, Cinzia Annunziata and Gennaro Casella

Obstetrics and Gynecology Unit, Villa Malta Hospital, Via Sarno Striano, 84087 Sarno, Salerno, Italy

***Corresponding author**
Anna Rita Palumbo, Obstetrics and Gynecology Unit, Villa Malta Hospital, Via Sarno Striano, 84087, Sarno, Salerno, Italy.

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Introduction

Gynandroblastoma is a rare ovarian tumor that manifests with a combination of two histological elements: granulosa cell tumor (GCT) and Sertoli-Leydig cell tumor (SLT) differentiation. (Jang NR 2018). Gynandroblastoma is a subtype of ovarian sex cord-stromal tumor and is usually observed in young women. The most important clinical manifestation is hormonal dysfunction [1]. Because this tumor is hormonally active, patients afflicted with it could present with either estrogenic or androgenic symptoms (Chivukula 2007). GCT presenting with pleural effusion has rarely been described in literature. We hereby report a patient with ovarian gynandroblastoma and pleural effusion [2,3].

Case Report

A 23-year-old nullipara woman presented with shortness of breath and episodes of pelvic pain. She had no history of medical or surgical diseases [4]. Radiographic images, ultrasonography (US), and computed tomography (CT) revealed right pleural effusions and a 24 cm x 25cm x 12 cm solid tumor in the right ovary containing cysts of various sizes.

- Abdominal and pelvic computed tomography (CT) revealed the presence of a large intra- abdominal formation, partly surrounded by fluid, measuring at least 26x21cm, probably originating from the uteroadnexal region, with a mixed supra-fluid/solid/sub-solid density which imprints and displaces the nearby organs and vascular structures, especially the delateralized colon leaning against the abdominal wall, the distal tract of the descending colon and the sigma displaced posteriorly.
- Abdominal and pelvic magnetic resonance (MRI) showed that: “most of the peritoneal cavity appears to be occupied by a large expanse (Dlong 25 cm, Dt 24 cm, Dap 12 cm) with a seromucinous signal and a multiloculated structure due to the presence of inhomogeneous impregnation after contrast medium”.
- Chest computed tomography (CT) manifested abundant right apical-parietal-basal pleural effusion with a maximum thickness of approximately 78 mm.
- Serum markers: carbohydrate antigen (CA)-125 was 175 IU/ml.

The patient underwent endopleural drainage placement with cytological and microbiological examination of the pleural fluid. The microscopic picture shows predominantly fluid amorphous material, blood cells and rare irrelevant mesothelial cells. In this context, a single group of cohesive elements with slight polymorphism was observed. Atypia of undetermined significance was also found.

The patient underwent right salpingo-oophorectomy, peritoneal washing, omental resection and multiple peritoneal biopsies [5]. Histopathological examination revealed microscopic and immunohistochemical findings, of complex interpretation, configuring an ovarian stromal tumor of the sexual cords with aspects that favor a possible diagnosis of adult granulosa cell tumor with multiple growth patterns and focal significant increase in the proliferative index [6]. Comment: the case described shows some atypical characteristics: the proliferative index focally higher than the average of adult granulosa cell tumors; also presence of a pseudoglandular pattern with focal positivity to cytokeratin AE1-3 and melan A that can however be positive in some stromal tumors including granulosa cell tumor

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(expression not constant nor specific). For the first consideration, a differential diagnosis with a juvenile form is required that seems to be excluded by the morphological aspects described. For the second note, the negativity to EMA and PAX8 allows us to exclude a neoplasm with endometrioid differentiation. However, the possibility of a differentiating component of Sertoli Leydig cells should be considered, which would also constitute an extremely rare neoplasm (gynandroblastoma) [7].

Literature

The present review has been conducted through systematic research of scientific publications about presentation of gynandroblastoma with pleural effusion. A systematic search of the literature was conducted using the keywords "gynandroblastoma" and "pleural effusion" to identify relevant studies reporting an association between this rare ovarian tumor and the presence of pleural fluid [8]. A total of 23 records were initially retrieved from electronic databases. After the removal of 2 articles that were not in English, 21 records were screened. No records were excluded at this stage. All 21 reports were retrieved and assessed for eligibility. Among these, 7 articles were excluded for being not relevant and 7 articles were excluded for involving other types of tumors. Finally, 7 studies met the inclusion criteria and were included in the systematic review. The PRISMA 2020 flow diagram (Figure 1) illustrates the study selection process. It clearly shows that the main exclusion occurred at the eligibility assessment stage, where non-relevant studies and those on different tumor types were removed. The absence of duplicate or non-retrievable records highlights the specificity of the search strategy [9,10].

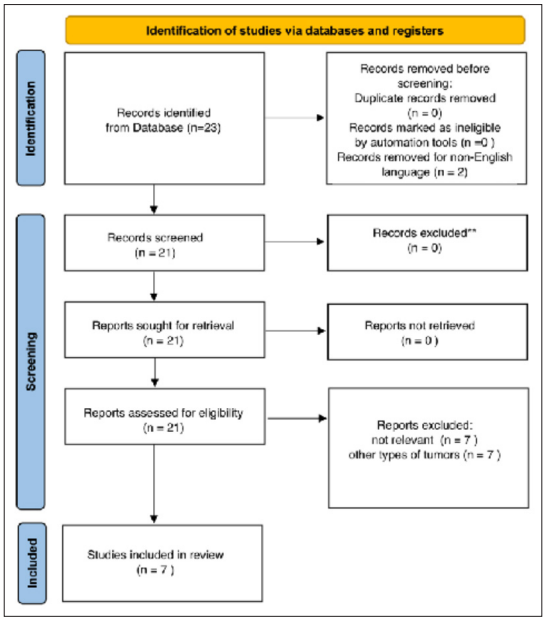

Figure 1: PRISMA 2020 Flow Diagram for New Systematic Reviews Which Included Searches of Databases and Registers Only

Discussion

In this systematic review, a total of 8 cases of gynandroblastoma associated with pleural effusion were analyzed (Tab 1.). The following key clinical and pathological aspects were evaluated:

Age at Diagnosis

The patients' ages ranged widely, from childhood (12 years old) to postmenopausal age (60+ years), with a median age leaning toward adulthood, suggesting that gynandroblastoma can occur across all reproductive phases. In the case we reported, the patient's age is the lowest compared to the rest of the population examined, with the exception of the cases of Kaur and Imai, aged 12 and 2 respectively, which instead report cases of juvenile granulosa cell carcinoma.

Elevated Tumor Markers

Several cases showed significant elevation in serum tumor markers. Estradiol levels were notably raised in multiple patients (up to 302 pg/mL), in 3 of the 7 studies analyzed from the literature.CA125 was analyzed in 4/7 studies, CA-125 levels were elevated in most cases (up to 708.69 U/mL). LDH was also elevated in some patients, suggesting a possible correlation with tumor activity or associated effusion.

Radiological Cystic Mass Dimensions

Radiologic imaging (ultrasound, CT, MRI) revealed the presence of adnexal masses, ranging in size from approximately 8.5 × 8.5 × 5.2 cm to 17 × 10 cm, often described as solid-cystic or heterogeneous. These findings indicate variability in the presentation but confirm the importance of imaging in initial tumor identification.

Malignant Cells in Pleural Effusion

Pleural fluid cytology was available in most cases. In some patients, malignant cells consistent with AGCT (adult granulosa cell tumor) were identified, while others showed inflammatory cells or were cytologically negative despite the presence of pleural effusion. This highlights that pleural involvement does not always imply cytologically detectable malignancy.

Conclusion

The present systematic review identified seven published cases of gynandroblastoma associated with pleural effusion, highlighting a rare but clinically relevant manifestation of this uncommon ovarian neoplasm [11]. The age range of patients spanned from adolescence to older adulthood, underscoring that gynandroblastoma- with or without pleural involvement- should be considered across a broad age spectrum. The frequent presence of elevated serum tumor markers (e.g., estradiol, CA-125, LDH) and adnexal masses of variable size suggests that early recognition of hormonal and radiologic abnormalities may facilitate prompt diagnosis [12].

From a therapeutic standpoint, most patients underwent total hysterectomy with bilateral salpingo-oophorectomy and additional staging procedures [13]. The management of the pleural component was heterogeneous; some patients received thoracentesis or pleural drainage, while the malignant nature of the effusion in others remained uncertain. Given the scarcity of cases, no standardized guidelines exist for pleural management in gynandroblastoma, but a multidisciplinary approach involving gynecologic oncology, thoracic oncology, and cytopathology is advisable [14,15].

In conclusion, gynandroblastoma complicated by pleural effusion is an exceptionally rare phenomenon, yet one with important clinical implications. Key take-home points include:

The occurrence of pleural effusion in these patients is notable. Although pleural involvement is more commonly associated with advanced gynecologic malignancies, the cases reviewed demonstrate that gynandroblastoma may also present with serous cavity spread or effusion, sometimes in the absence of other obvious metastatic findings [16-18]. Cytological examination of pleural fluid revealed malignant cells in a minority of cases; more often effusions were described as exudative but without direct cytologic confirmation of tumor cells.[19]

Radiologic assessment revealed adnexal masses ranging from approximately 8.5 cm to nearly 17 cm in diameter, often described as solid-cystic with variable features. These findings emphasize the importance of imaging in the initial work-up of gynandroblastoma [20-22]. However, the presence of pleural effusion should prompt thoracic imaging and cytologic/histologic evaluation of the fluid and pleura, especially when accompanied by thoracic symptoms or elevated tumor markers [23].

- In women presenting with adnexal masses, elevated serum tumor markers, and unexplained pleural effusion, gynandroblastoma should be included in the differential diagnosis.
- Imaging of both the pelvis and thorax, combined with cytology and immunohistochemistry of pleural fluid or tissue, is essential for accurate staging and management.
- Surgery remains the mainstay of treatment, but pleural involvement calls for a collaborative thoracic-gynecologic oncology strategy.

Further accumulation of individual case data and longer-term follow-up are needed to better define optimal management strategies and prognostic factors in this rare setting. Researchers and clinicians are encouraged to report new cases of gynandroblastoma with pleural involvement to improve the evidence base and guide future therapeutic recommendations [24,25].

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