

Primary Ovarian Mesothelioma: About a Rare Case

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To cite this article:

Adadi Hind, Fdili Alaoui Fatima-zahra, Jayi Sofia, Chaara Hekmat, Melhouf Moulay Abdelilah. Primary Ovarian Mesothelioma: About a Rare Case. *World Journal of Medical Case Reports*. Vol. 3, No. 1, 2022, pp. 5-7. doi: 10.11648/j.wjmcr.20220301.12

Received: November 23, 2021; **Accepted:** January 5, 2022; **Published:** January 20, 2022

Abstract: Malignant Mesothelioma of Ovary is an extremely uncommon, malignant tumor of the ovary. The cause of formation of the tumor is not well understood. The clinical presentation of MMO is equivalent to Large-sized tumors that causes abdominal and pelvic pain, discomfort, and weight loss. The complications of Ovarian Malignant Mesothelioma (OMM) include infertility, metastasis to various regions, and recurrence following its surgical removal. It's prognosis is generally poor due to local invasion and metastasis of the malignancy to various body site. Here we report the case of a 50-year-old patient presenting with an increase in abdominal volume associated with a deterioration in general condition. The abdomino-pelvic CT showed a huge left ovarian without peritoneal carcinoma. The final pathologic diagnosis of ovarian malignant mesothelioma was made through histologic and immunohistochemical examination. The surgery was followed by adjuvant chemotherapy, resulting in a complete radiological response, with disease-free survival of more than one year.

Keywords: Primary Ovarian Mesothelioma, Immunohistochemistry, Uncertain Prognosis

1. Introduction

Malignant mesotheliomas (MM) are very aggressive primary tumors. They mainly occur in the pleural, peritoneum, pericardium or in the tunica vagina. Malignant ovarian mesothelioma (MMO) is a very rare tumor, very little described in the literature, so the knowledge of its natural history is very limited. Given the limited number of cases reported in the literature, this is a tumor that remains poorly characterized epidemiologically, clinically, radiologically and histologically. Here we report a case of MMPO: Its clinical characteristics, detailed radiological and histopathological results, as well as therapeutic management will be discussed.

2. Case Presentation

2.1. Clinical History

This is a 50-year-old Moroccan woman, menopausal, with a history of left heart failure who was admitted to the hospital with lower abdominal pain and an increase in abdominal volume associated with a deterioration in general condition for about six months. She had no known asbestos exposure. Clinical examination revealed a left adnexal mass. On

examination, her vital signs and CA 125, carcinoembryonic antigen tumor serum markers and the CA 19-9 were within normal ranges. Transvaginal ultrasonography showed a 20 × 13 × 11 cm cyst with solid component. The abdomino-pelvic CT demonstrated a voluminous cyst (20 × 10 × 09 cm) and the presence of ascites. We performed a laparotomy that showed a mass measuring 20cm * 13cm, it was on her left ovary, containing bloody fluid. In her pelvis we can see the encapsulated bloody effusion, which was adhered to omentum. In the other hand the uterus and the right ovary were normal. We did a total hysterectomy with bilateral adnexectomy and multiple biopsies. There was no nodules on the liver, peritoneum, and other abdominal sites on gross inspection in surgery.

2.2. Pathological Findings and Immunohisto Chemistry

1. Macroscopic examination: left ovarian mass 22x11x10 cm. When cut, we noted the result of a hemorrhagic fluid with a heterogeneous appearance and presence of fleshy areas and other cysts.
2. Microscopic examination: we noted an undifferentiated tumor proliferation of diffuse and papillary architecture. Tumor cells are increased in size and very atypical with prominent nucleoli, abundant eosinophilic cytoplasm,

and numerous mitoses.

- Immunohistochemical study: (after a large panel with negative antibodies) we noted a diffuse expression of CK, CK7, Calretinin, Vimentin and WT1 and focal of Inhibin, CK8 / 18, CK20 and the CK19.

We have concluded that there is a carcinomatous process whose immunohistochemical profile points towards ovarian mesothelioma.

In our case, the patient was referred to oncology for chemotherapy then we lost contact with her.

3. Discussion

MM is a tumor derived from mesothelial cells. Exposure to asbestos is the most common cause [1]. However, several cases in the literature were not associated with this notion [1]. Our patient had a negative history of occupational exposure to asbestos, as she is a housewife. MM can occur after radiation therapy, suggesting that direct radiation may be a risk factor for its development [1, 4]. In this case, there is no notion of radiotherapy. Other factors have been implicated including certain minerals, such as erionite, thorium and mica [1, 5]. However, causality related to these substances has only been reported in isolated cases and therefore the relative risk of developing MM has not yet been quantified [1, 11].

Primary ovarian mesothelioma accounts for approximately 0.03% of mesothelioma-related deaths [6, 10]. Ovarian localization is frequent when there is diffuse or multifocal involvement of the peritoneum. In addition, the literature has reported only a few cases of MM presenting mainly with an ovarian mass [2]. Goldblum and Hart had found secondary ovarian disease in 10 cases during diffuse peritoneal mesothelioma; the mean age of the cohort was 52 years [7]. Clement *et al.* reported a series of 9 cases of MM presenting as ovarian masses. The mean age of the patients was 52 years; 7 of the 9 presented with an extensive extraovarian peritoneal tumor, while the tumors were confined to the ovaries in 2 patients [8]. In her series of 7 mesothelioma cases, Merino had patients between 22 and 52 years old with an average age of 32 years [6]. Our patient is 50 years old, which goes back to the literature. The most frequently reported first symptoms are abdominal pain (35%), with abdominal distension (31%), anorexia, weight loss and ascites [2].

Sometimes the diagnosis is made incidentally during a laparoscopy [2]. In immunohistochemistry, malignant mesothelioma is characterized by the positivity of calretinin, cytokeratin 5/6, EMA (Epithelial Membrane Antigen), Wt1 (Wilms' tumor gene 1), cellular anti-mesothelial antibody 1 and mesothelin. In addition, we note the negativity of markers of malignant tumors of the gastrointestinal tract including ACE and MOC-31 (or B72.3, Ber-EP4 or BG-8) [2]. For our patient, the immunohistochemistry showed positivity for cytokeratin 7, calretinin, vimentin, while we have the negativity of cytokeratin 5/6, EMA, and Wt1. Ber-EP4 was weakly positive. Ovarian adenocarcinoma and malignant mesothelioma both co-express cytokeratin, vimentin, and often EMA. In another work, only Ber-EP4,

MOC-31, the estrogen receptor and calretinin were retained as useful markers for this differential diagnosis [2]. Overall, it is recommended to use at least two markers for mesothelioma and two markers for carcinoma [9, 13]. In the ovary, mesothelioma must be differentiated from other neoplasms, including benign lesions such as cystic lymphangioma, cystic forms of endosalpingiosis, endometriosis and cystic adenomatoid tumors [6].

The therapeutic strategy is based on a locoregional approach by optimal or suboptimal cytoreduction surgery, which increases the chances of long-term survival in these patients. First-line systemic chemotherapy is reasonable for tumors that are unresectable at first, but the response is limited. This chemotherapy also finds its place in an adjuvant situation in the presence of factors of poor prognosis. The active drugs that have been used in the various cases reported are: platinum salts (Cisplatin, Carboplatin), Gemcitabine and Pemetrexed. Historically, chemotherapy regimens have been Irinotecan plus Carboplatin, ideally Cisplatin 50 mg / m² plus Pemetrexed 500 mg / m². Intra-peritoneal chemotherapy with various agents including Cisplatin, Mitomycin C, Doxorubicin, Epirubicin, Etoposide and Cytarabine, used alone or in combination has also been tested [2, 8, 12].

4. Conclusion

Primary involvement of the ovaries by malignant mesothelioma is exceptional and only a few cases have been reported in the literature. Differentiation occurs with diffuse peritoneal mesothelioma, ovarian carcinoma and peritoneal carcinoma. New immunohistochemical markers such as calretinin confirm the diagnosis. The therapeutic management is multidisciplinary based on surgery, chemotherapy and radiotherapy, but more effective strategies are necessary in order to improve the results of conventional approaches. Further studies are required for early and correct diagnosis. Knowledge of this presentation is important in order to establish appropriate surgical and adjuvant therapies.

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