

Ruptured ovarian metastatic malignant melanoma caused acute abdomen

Case report

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Abstract: Ovarian metastatic malignant melanoma is a rare form of disseminated malignant melanoma. We present a rare case of acute abdomen due to rupture of ovarian metastatic malignant melanoma seven years after removal of a primary cutaneous malignant melanoma lesion, followed by reexcision of the cicatrix and axillary dissection (Clark III, Breslow IV), one year after osteoplastic parietal craniotomy for removal of recidiv metastatic lesions, and excision of the cutaneous malignant melanoma lesion on the upper leg were performed. During laparotomy because of acute abdomen, 4 L of free liquid (blood and ascites) were evacuated. The right adnexal mass was loose tumor, size 110x75 mm, with rupture on the posterior wall and hemorrhage. Unilateral adnexectomy was performed. Pathohistologic evaluation revealed tumor cells with eosinophilic, clear cytoplasm, intracytoplasmatic melanotic pigment and a great number of mitosis. Immunohistochemical results supported positivity for protein S-100, whereas results for cytokeratin 7, cytokeratin 20, pancytokeratin, epithelial membrane antigen and HMB-45 were negative. Three months after the surgery the patient died due to disseminated cerebral melanoma. An adnexal mass and the history of previous MM should be suspected to be ovarian metastatic malignant melanoma and the patient should be seen by gynecologist at least for active treatment.

Keywords: *Melanoma malignum* • *Metastasis* • *Ovarium* • *Acute abdomen*

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1. Introduction

Malignant melanoma (MM) is a very malignant tumor with an extremely high metastatic potential and an unpredictable profile of spread. The incidence of cutaneous malignant melanoma is increasing at an alarming rate around the world for decades in fair-skinned, indoor-working populations, and may be increasing at an exponential rate [1].

Metastasis of the MM in the genital tract are rare tumours. Ovarian metastatic malignant melanoma (OMMM) is a rare form of disseminated malignant melanoma. It has unpredictable clinical and biological behavior and bad prognosis due to the aggressiveness of the tumor. OMMM presenting initially as an ovarian tumor is uncommon and causes diagnostic conundrums, especially after long periods of remission [2].

We present a rare case of acute abdomen due to the rupture of OMMM seven years after removal of a cutaneous malignant melanoma lesion, followed by reexcision of the cicatrix and axillary dissection. Acute abdomen occurred a year after osteoplastic parietal craniotomy for removal of the metastatic lesions and excision of the cutaneous malignant melanoma lesion on the upper leg.

2. Case report

A 35-year-old patient, hairdresser, mother of two children, was admitted to the emergency unit with abdominal pain, pelvic mass, profuse sweating, nausea and vomiting. Those symptoms started two days before admittance. Her medical history: at the age of 16 she

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underwent cystectomy of the left ovary (histological examination showed benign cyst). Familial history was without abnormalities. Seven years ago she underwent excision of primary cutaneous malignant melanoma lesion located in interscapular region. Histological finding was: melanoma malignum (MM), Clark III, Breslow IV.

Three weeks after primary surgery, reexcision and axillary dissection was performed; histological examination found no sign of metastasis. At regular medical check up one year ago, solid brain metastasis was diagnosed. Therefore she received 3 cycles of monochemotherapy (dacarbazine). After chemotherapy she underwent osteoplastic parietal craniotomy with ablation of metastatic lesion in the frontal cerebral lobe. Metastasis of MM was histologically confirmed. Palliative radiation therapy was administered to the patient and followed with 3 more cycles of chemotherapy (dacarbazine). 4 months after brain surgery, excision of cutaneous MM lesion was performed, histological finding showed metastatic nodular melanoma 3.5 cm in depth. Just before admittance in our hospital, she received 4 cycles of second line of chemotherapy (protocol DBD-doxorubicine, bleomycin, dacarbazine). A month before admittance she was referred to bone scintigraphy that showed vertebral metastasis—oval aggregation in L4 trunk. MSCT scan revealed ascites in abdomen, a uterus of normal size, a right adnexal solid cystic mass 8x5x6.5 cm, which was partially fluid filled in the cystic part. The liver and spleen were enlarged, but without any sign of metastases. Chest X-ray showed no abnormalities. Ultrasound showed ascites and solid cystic tumor of the right ovary measuring 100x75 mm with detectable central and peripheral vascularisation. Doppler analysis of tumoral blood flow revealed doppler resistance index (RI) of 0.24-0.38.

After admittance, her CBC (complete blood count) showed erythrocytes (E) 2.2, hemoglobin (Hgb) 7,4 g/dl, hematocrit (Htc) 21,7 %. On the second day she developed a significant decrease in RBC (red blood count): E 1.84, Hgb 6,3 g/dl, Htc 17,8 %, but without any signs of acute abdomen or hemorrhagic shock. Due to anaemia she received a blood transfusion after which an increase of RBC was noted (E 3.06, Hgb 98,0, Htc 29 %). As she was complaining about back pain and abdominal pain, she received a fentanyl patch. 9 days after admittance she was in a poor condition. The patient suffered from diffuse abdominal pain and nausea, and subsequently developed peritonitis. According to CBC she was anaemic again: E 1.93, Hgb 6,0 Htc 18,3 % followed by hyperfibrinogenemia (9.2 g/L, D-dimer 1332, PT 0.70, APTT 37.1) and thrombocytosis. Ultrasonography detected opalescence of free abdominal liquid. Due to the progression of anaemia and the development of an acute

abdomen, diagnostic laparocentesis was performed and blood received, BP 120/80 mmHg, pulse 140/min. Suspicion of adnexal mass rupture causing acute abdomen was the reason for exploratory laparotomy. Indication for surgery and possible complications were discussed with the patient and her family, and they agreed that immediate surgery should be performed. The patient underwent lower midline laparotomy under general anesthesia. After opening of the abdominal cavity, 4 L of free liquid (blood and ascites) were evacuated. The right adnexal mass was loose tumor size 110x75 mm with rupture on the posterior wall and hemorrhage. Unilateral adnexectomy was performed. Exploration of abdominal cavity found no macroscopic signs of metastases. The uterus and left ovary were without any abnormalities.

During the surgery, the patient was hypotensive (BP 100-110/50-50 mmHg) with tachycardia and received 1480 mL of erythrocyte transfusion and 760 mL of fresh frozen plasma with perioperative antibiotic prophylaxis (cephazolin). CBC after the surgery and completed transfusion showed improvement: E 2.81-3.79, Hgb 8,6-11,4; Htc 25-34 %, Plts 334- 480, and renal and hepatic function were intact. Postoperative care and recovery period were normal and the patient was continuously receiving low-molecular-weight heparin for thromboprophylaxis. 10 days after the surgery, the patient was discharged in good physical condition. The tumor was partially necrotic, well vascularised and consisted mostly of solid parts. Microscopic evaluation revealed tumor cells with eosinophilic, clear cytoplasm, intracytoplasmic melanotic pigment and a great number of mitosis. Immunohistochemical results supported positivity for protein S-100, whereas results for cytokeratin 7, cytokeratin 20, pancytokeratin, epithelial membrane antigen and HMB-45 were negative. These findings confirmed that the adnexal tumor was OMMM. Postoperative control a month after the surgery found no abnormalities.

Three months after the surgery patient died due to disseminated cerebral melanoma.

3. Discussion

MM can disseminate in the brain, liver, vagina, perineum, lymph nodes, ovarium, skin etc. OMMM are very rare tumors and primary ovarian melanoma (OMM) is extremely rare [3]. These tumors are rarely predictable and are being diagnosed in the advanced stage of the disease.

Although persons of any age may be affected, the peak incidence is in the reproductive age. Ultrasound, MRI and MSCT scan revealed an adnexal mass, mostly

unilateral. OMMM usually appears as a solid ovarian mass, rarely as a solid cystic or multicystic mass. These neoplasms are often difficult to differentiate from many other types of tumors, including dermoid cysts, cystadenoma mucinosum, or juvenile granulosa cell tumor and small cell carcinoma because of the presence of follicle like spaces. Ascites occurs in approximately one third of the patients [4-6]. Postmortal diagnosis of OMMM is conducted in approximately 20% of the cases. Diagnosis should be done according to immunohistochemical results [7]. Ueng *et al.* have published a study about primary OMM in 4 cases, and 1 case of OMM inside of a teratoma [8]. The incidence of primary cutaneous malignant melanoma is proportionate to tumor depth if the depth of the lesion is > 0.76 mm. In all of primary OMM described cases with primary cutaneous lesions, there is a poor five-year survival rate for stage III (45%) and stage IV (11%) of the disease [9].

Until year 1998, 73 cases of OMMM and 20 cases of OMM have been published [7]. Gupta and coworkers have found all together 23 cases of OMM in 40 years of clinical data [4]. They found 19 unilateral and 4 bilateral OMM that measured 4.5-23 cm in diameter (average 10 cm). Altogether they reported about 35% of pigmentous MM lesions. According to the study, just two patients survived, their disease free survival was 24 and 96 months.

Sabban *et al.* published a case report about a 31-year-old patient admitted to the hospital due to pelvic pain and an ovarian cyst 9 cm in diameter [10]. The patient had a history of cutaneous MM that was operated 5 years ago. Adnexectomy was performed, and histological examination combined with immunohistochemistry (protein S-100 positive as well as HMB-45, cytokeratin negative) confirmed diagnosis of OMMM.

A very interesting case was reported by Oliver *et al.* [2]. A 58-year-old postmenopausal patient was incidentally found to have an abdominal mass, 10 years after excision of a cutaneous MM lesion (Breslow 1.4 mm, Clark's level III without vascular invasion) in the cubital region. At the time of admittance she presented with ascites and a multilocular cyst-Omm that measured 8x6x12 cm. The patient was operated; total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH/BSO), lymphadenectomy and infracolic omentectomy were performed. After the surgery her condition rapidly deteriorated and postmortal examination showed that at the time of death disease presented with metastases to brain, lungs and pancreas.

Complications of ovarian tumor such necrosis, torsion, rupture or infiltration are well know, but according to the data from literature, there is not a single report about acute abdomen due to a rupture of OMMM. One

case of acute peritonitis due to unilateral OMMM rupture was described by Silveira *et al.*, in 1977. [11]. Jaluvka *et al.* [12] presented a case of a 26-year-old who presented

with lower abdominal pain. 10 years ago she underwent excision of superficially spreading MM from the left temple. On examination, cystic adnexal masses were detected and an operation was indicated. Intraoperatively bilateral polycystic, dark-colored ovarian tumors were found, each measuring 10x10x10 cm. TAH/BSO and resection of the great omentum was performed. The uterus and ovaries weighed 3480 g and histological examination revealed diagnosis of pigmented melanoblastoma in both ovaries. Two months after surgery, the patient succumbed to multiple cerebral metastases. A similar case of choroidal melanoma which metastasized to the ovary was reported Mandato *et al.* [13] and OMMM with omental infiltration 25 years after the primary surgery of choroidal melanoma was described by Ben David *et al.* [14].

Moselhi *et al.* published 3 cases of OMMM with previously excised cutaneous lesions. The first patient underwent excision of the cutaneous lesion located on the upper leg 8 years ago (Clark 3, Breslow 2 mm). The second patient had a similarly located MM that was removed 2 years ago (Clark 3, Breslow 1 mm), and the third was operated on due to MM located on the shoulder (Clark 3, Breslow 1 mm). All 3 of them were in the reproductive age (36, 25 and 45 years). All of them presented with pelvic masses: solid ovarian tumor 10x10 cm in greatest diameter, cystic mass 5 cm in range and complex adnexal mass 8 cm in greatest diameter accompanied by ascites. Chemotherapy was administrated after the surgery to the first two described patients; unfortunately they died due to dissemination of the disease. The third patient was not operated, and diagnosis of amelanotic MM was confirmed by cytological examination of ascites obtained by laparocentesis [5]. Piura *et al.* described a case of OMMM found in a 45-year-old woman who underwent surgical removal of the primary cutaneous lesion (Clark III, Breslow 0.82 mm) on her back 7 years ago. She presented with a left adnexal mass measuring 10x7x7 cm. The patient had TAH/BSO, infracolic omentectomy and selective retroperitoneal lymphadenectomy, followed by whole brain irradiation and chemoimmunotherapy. Histology and immunohistochemistry confirmed diagnosis (protein S-100 and HMB-45 highly positive, cytokeratin negative). Seven months after the OMMM was diagnosed, the patient succumbed to multiple cerebral metastases and metastasis to the skin of the axilla [7].

We described a typical clinical course of the disease with a primary cutaneous MM lesion, followed by relapse after a prolonged period of remission, presenting

as an isolated ovarian metastasis of MM. She developed acute abdomen due to the rupture of the ovarian cyst, complicated by intra-abdominal hemorrhage. Due to vital indication, urgent exploratory laparotomy and adnexectomy were performed.

This case is among the rare described cases of OMMM, but among them no case described a case of acute abdomen due to the rupture of OMMM. Adnexal mass and the history of MM should be suspected to be

OMMM and the patient should be seen by a gynecologist at least for active treatment.

Also, for surgeons, if they see MM patients with abdominal pain, there is a possibility of acute abdomen due to the rupture of the lesion. Aggressiveness of MM and its unpredictable biological behavior often results in disseminated disease that is not curable, neither by surgery nor by chemoirradiation.

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