LATE RECURRENT OVARIAN CARCINOMA METASTATIC TO THE THORACIC WALL

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ABSTRACT

Ovarian cancer is the second most common gynaecologic malignancy. Ovarian carcinomas typically metastasize to multiple sites via exfoliation, lymphatic spread or direct invasion. We present a rare case of a very late recurrence of ovarian carcinoma into the thoracic wall, heralded by thoracic pain in a patient otherwise disease-free for 23 years. This unusual and late presentation of an ovarian cancer metastasis underscores the need for continued awareness and attention to new symptoms in patients with ovarian cancer who show prolonged disease-free intervals.

Key words: pleural mass, radiotherapy, thoracic surgery

INTRODUCTION

Ovarian cancer is associated with an overall recurrence rate of 50 to 75% of responders after completing first-line therapy (1). Very late relapse of ovarian cancer is unusual and may present with atypical symptoms. We describe a case of very late recurrence of an ovarian carcinoma, 23 years following initial surgical therapy, and occurring at an unusual location, the thoracic wall.

CASE REPORT

A 59-year-old woman presented in September 2007 at the Pulmonology department with a sudden feeling of a painful lump at the back, located caudally of the right scapula. One year prior to presentation, she developed an atypical bilateral thoracic pain and increased dyspnoea on exertion. She had a history of appendicitis in 1962. In 1978, she underwent unilateral right salpingo-oophorectomy for an unknown reason. In 1984, she was diagnosed with ovarian cancer and treated with total abdominal hysterectomy with left salpingo-oophorectomy and adjuvant chemotherapy. The pathology examination established the histological diagnosis of a serous-papillary ovarian carcinoma with small implantations at the peritoneal surface and the diaphragm. The patient had no smoking history.

At present, the ultrasound examination showed a large (9.8 cm diameter) intra-thoracic mass with both cystic and soft tissue components. A Computed Tomography of her chest confirmed the presence of this intra-thoracic mass, but also showed evidence of regional invasion into the intercostal muscles, the vertebral body, the ribs and the posterior mediastinum (Figure 1A).

A whole body 18-fluoro-2-deoxyglucose Positron Emission Tomography (FDG-PET) scan showed increased focal tracer uptake at the posterobasal pleura of the right lung (Figure 1B). The serum marker CA 125 was increased: 270 kU/L (normal value below 35 kU/L). The patient first underwent two CT-guided transthoracic biopsies, which were non-diagnostic and, thereafter, a surgical biopsy was performed. Histopathological examination of the resected specimen demonstrated malignant cells lying in a columnar epithelial layer with focal papillary growth. Immunohistochemical staining was positive for CEA and negative for calretinin and TTF1, confirming the pathological diagnosis of a metastasis of a serous-papillary ovarian carcinoma (Figure 1C). Because of the slow growth pattern of the tumour, and therefore presumed weak sensitivity to chemotherapy, a surgical excision of the tumour with thoracic wall resection was planned although a complete resection could not be guaranteed based on the CAT findings. A resection “en bloc” of the right posterobasal thoracic wall including ribs 9-12, part of the diaphragm, the azygos vein, the thoracic duct, the sympathetic chain, and the mediastinal fat was performed. A complete right lower lobectomy was necessary for technical reasons because the tumour deeply invaded the lung parenchyma over a large area of the circumference. The chest...
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The wall was reconstructed with an non-absorbable polypropylene prothesis (Marlex®). A mediastinal lymph node dissection was carried out. Several lymph nodes (9 out of 11 resected nodes) in the hilar and intralobar region, but also the lymphatic vessels along the thoracic duct into the neck were found positive and these vessels could not be completely removed at the time of the operation. The procedure was complicated by a chylous leak necessitating surgical re-intervention on postoperative day 30 with ligation of the thoracic duct. No adjuvant chemotherapy was given, but the patient received local radiotherapy to the mediastinum and the posterior thoracic wall (fractions of 2 Gray with a total radiation dose of 50 Gray). Two months after ending radiotherapy, this patient still presented with some right upper abdominal pain, but there was no radiological proof of local tumour relapse and the CA 125 tumour marker normalized (26 kU/L). One year after ending radiotherapy, the

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Figure 1: A: CT scan of thoracic wall metastasis shows extensive regional tumoural invasion [marked with arrows]. B: FDG-PET scan of the thorax demonstrates focal FDG tracer uptake [marked with arrows] in the posterobasal region of the right hemithorax. C: Pathological findings in the surgical biopsy of the intrathoracic mass. 1: Low power view shows a papillary tumour at the surface of a thickened fibrous pleura [* denotes pleura] (H&E X 25). Field of magnification shown in Figure 2 has been marked. 2: The fibrovascular papillary structures are covered by tall cylindrical and fairly atypical epithelial cells. Their immunohistochemical profile was inconsistent with mesothelial cells (H&E X 200).
patient did not present any new symptoms. The CA 125 marker level was slightly elevated (40 kU/L) and on a whole-body PET-CT scan, three new, but small pleural nodules were detected in the right hemithorax. A watchful waiting approach was advised at first.

**DISCUSSION**

In Europe and in the United States of America, ovarian carcinoma is the most common cause of death among women with gynaecologic malignancies. The disease usually remains confined to the peritoneal cavity at presentation and throughout its further course. Although the intraperitoneal route of dissemination is the most common, ovarian cancer may also metastasize through the lymphatic channels and the haematogenous route. Distant metastases may occur at the time of ovarian cancer diagnosis (stage IV disease) or may arise during the course of the disease. A 5-year large retrospective study found that 44% of the patients had metastasised to the thorax, mainly involving the lung parenchyma or pleura (2).

Another retrospective analysis in 73 ovarian cancer patients showed that 37 of them (52%) disclosed some kind of thoracic involvement in autopsy evaluation, sometimes remaining undiagnosed prior to death (3). However, thoracic wall involvement is an infrequent finding in ovarian cancer. To our knowledge, only three case reports have been published describing thoracic wall metastases of ovarian cancer, occurring between 30 months and 8 years after surgery for the primary tumour (4).

In our patient, the time to recurrence, 23 years after the diagnosis, was unusually long. A retrospective study including 400 patients with ovarian cancer showed that 87% of relapses occurred within 5 years following primary treatment, 8% relapsed between 5-10 years and 6% relapsed after 10 years (5). Very late relapses, after more than 20 years have only rarely been reported for invasive ovarian cancer. In all the described cases of relapse occurring after 20 years, abdominal involvement was present. In contrast, in our patient, only thoracic wall involvement was present, without the presence of peritoneal lesions, was disclosed after more than 20 years of follow-up.

Curative treatment of recurrent disease is unlikely. Hence, the goal of treatment of recurrent disease is disease control taking quality of life into consideration. A majority of recurrences develop within one year and second-line chemotherapy rather than surgery is indicated in symptomatic or life-threatening disease. In ovarian cancer recurring more than 1 year after primary treatment, secondary debulking may be indicated in selected patients. If the disease is localized, a good performance status is present and response to first-line therapy occurred, secondary debulking surgery followed by combination chemotherapy is advised (6).

The current case is exceptional in time to recurrence and localization of recurrence. Given the long disease-free interval and localized lesion, we opted for a radical surgical resection. Surgical removal was opted for since a long disease-free interval suggests an indolent growth that is likely to be less responsive to chemotherapy. This case illustrates that biopsy of any mass in patients with a history of ovarian cancer is mandatory, especially when the interval is long. Treatment for recurrent disease should be individualized. A multidisciplinary approach is advised to assess the benefit and potential to remove an isolated distant relapse.

One year after radiotherapy, the CA 125 marker level was slightly elevated and three pleural nodules were detected on PET-CT scan. Because a recent randomised trial showed no difference in survival and quality of life when asymptomatic ovarian cancer patients with elevated CA 125 level were treated for their relapse immediately or only when they developed symptoms, a watchful waiting approach was advised (7).

**CONCLUSION**

This case demonstrates that very late relapses of ovarian cancer may occur also at an unusual location, the thoracic wall. The metastatic thoracic wall lesion in our patient was discovered only because of the resulting thoracic pain, and highlights the need for close scrutiny of new symptoms in patients with ovarian cancer who experience prolonged disease-free intervals.

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**REFERENCES**