Taiwanese Journal of Obstetrics & Gynecology 60 (2021) 136–138

Contents lists available at ScienceDirect

Taiwanese Journal of Obstetrics & Gynecology

journal homepage: www.tjog-online.com

Case Report

Ovarian mucinous cystadenoma with a mural nodule of osteosarcoma: A case report

Ling-Hui Chu [a], Hung-Cheng Lai [a, b], Yu-Tso Liao [c, *]

[a] Department of Obstetrics and Gynecology, Shuang Ho Hospital, Taipei Medical University, New Taipei City, Taiwan
[b] Department of Obstetrics and Gynecology, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan
[c] Department of Colorectal Surgery, National Taiwan University Hospital Hsin-Chu Branch, Hsin-Chu, Taiwan

ARTICLE INFO

Article history:
Accepted 20 February 2020

Keywords:
Mucinous cystadenoma
Osteosarcoma
Ovarian cancer
Ovary

ABSTRACT

Objective: Osteosarcoma as a mural nodule in the ovary is extremely rare. We aimed to describe a case of a mural nodule with features of an osteosarcoma arising in an ovarian mucinous cystadenoma.
Case report: The 65-year-old woman presented with progressive abdominal swelling and poor intake. Image studies showed a huge (diameter, >30 cm) intra-abdominal multiloculated cystic lesion, suspected to be an ovarian tumor. She underwent unilateral salpingo-oophorectomy with no postoperative adjuvant therapy. She was disease-free at 16-month follow-up.
Conclusion: Osteosarcoma presenting as a primary ovarian neoplasm is rare, either as a pure osteosarcoma or arising from a teratoma. However, two osteosarcoma cases occurring arising from a mural nodule in an ovarian mucinous neoplasm have been reported. There is no consensus regarding the treatment strategy for osteosarcomatous mural nodules in mucinous tumors because of its rarity. More case studies are needed before its pathogenesis can be fully understood.

Introduction

Solid mural nodules are not commonly found in ovarian mucinous neoplasms of intestinal type. In 1979, Prat and Scully first described ovarian mucinous tumors with sarcoma-like mural nodules as possibly reactive lesions mimicking sarcoma [1]. Reviewing the literature, rare cases of ovarian mucinous tumors were associated with mural nodules which had either benign histology, such as leiomyoma, rhabdomyoma, or hemangioma, or malignant histology, such as anaplastic carcinoma, sarcoma of various types (mostly undifferentiated), or carcinosarcoma [1,2] (see Table 1).

Osteosarcoma arising from a mural nodule in the ovary is extremely rare. Only two cases have been previously reported [3]. We aimed to describe a case of a mural nodule with features of an osteosarcoma arising from an ovarian mucinous cystadenoma.

Case report

History and investigation

A 65 year-old woman underwent total vaginal hysterectomy because of uterine prolapse. She presented with progressive abdominal swelling which she had experienced for the previous 2 years. She visited our hospital due to concomitant poor intake. Abdominal ultrasound revealed a huge cystic lesion in the abdominal cavity. Abdominal and pelvic computed tomography showed a huge (diameter, >30 cm) intra-abdominal multiloculated cystic lesion. Ovarian tumor was considered. Her tumor markers showed mildly elevated CA19-9 (41.506 U/ml) and normal CA125 (23.5 U/ml) and CEA (0.438 ng/ml).

The patient underwent right salpingo-oophorectomy and enterolysis. She had an uneventful recovery and was discharged 4 days after surgery. The tumor was staged as International Federation of Gynecology and Obstetrics (FIGO) IC, and she refused further chemotherapy for personal reasons. She was followed up without adjuvant therapy for 16 months. The patient did not exhibit any evidence of tumor recurrence.

* Corresponding author. Department of Colorectal surgery, National Taiwan University Hospital Hsin-Chu Branch, No. 25, Ln. 442, Sec. 1, Jingguo Rd., North Dist., Hsinchu City, 300, Taiwan. Fax: +886 223934358.
E-mail address: yutsoliao@gmail.com (Y.-T. Liao).

https://doi.org/10.1016/j.tjog.2020.10.002

Table 1
Clinical features, therapy, and outcome of patients with osteosarcoma as mural nodules in ovarian mucinous tumors.

Case no.	Age	Presenting symptoms	Stage	Size	*Treatment*	Adjuvant treatment	Follow-up
1. McFarland, 2015	34	Persistent vaginal bleeding & abdominal swelling	IA	29 cm	TAH, bilateral SO, Oment	–	18 m NED
2. McFarland, 2015	18	Abdominal swelling	IC	NS	Left SO, Oment	–	12 m NED
3. Present report	65	Abdominal swelling	IC	>30 cm	Right SO	–	5 m NED

NED: no evidence of disease.
DOD: dead of disease.
SO: salpingo-oophorectomy.
Oment: omentectomy.
NS: not stated.

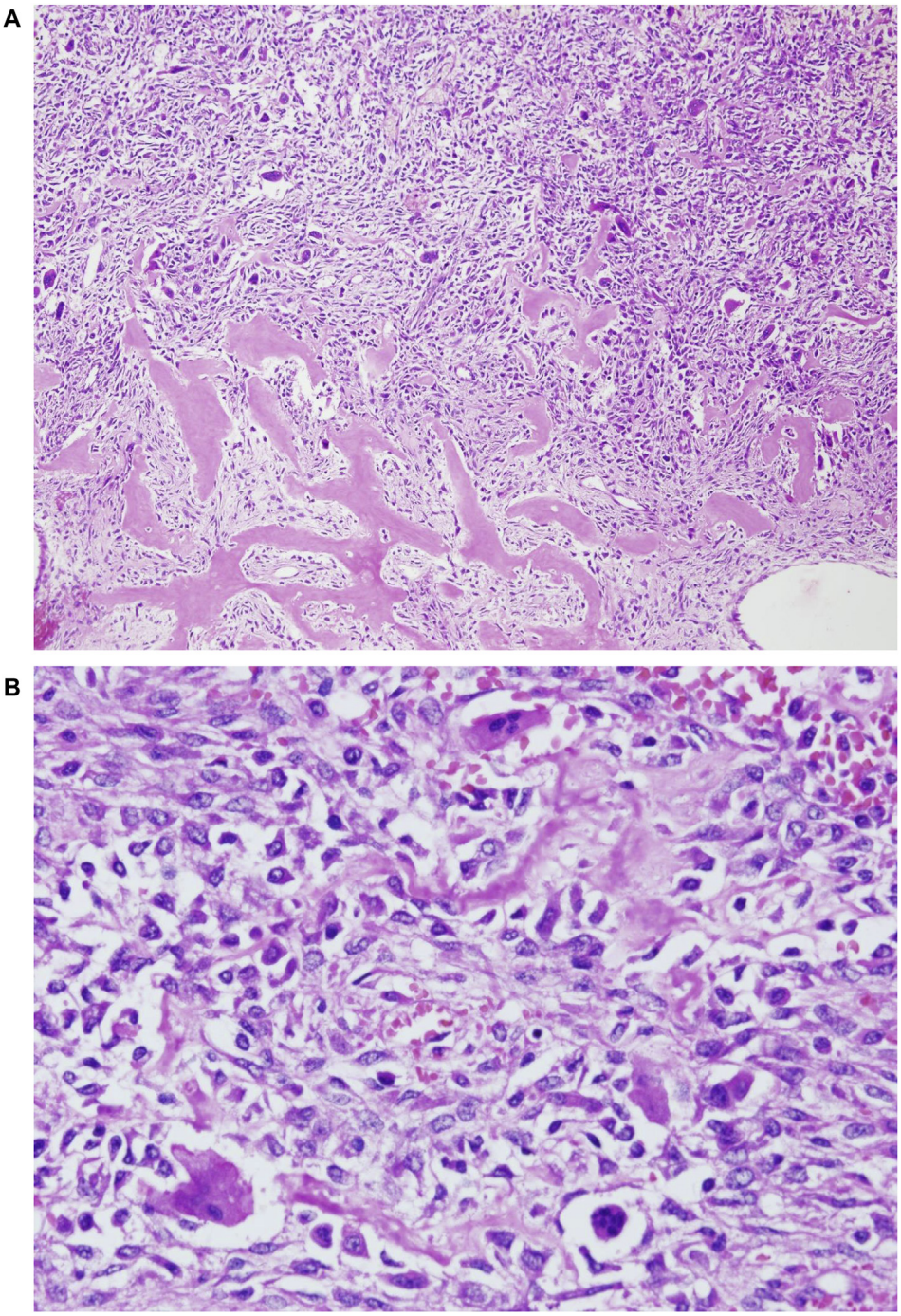

Fig. 1. (A) The mural nodule is composed of several mononuclear cells and multinucleated giant cells. Increased osteoid matrix formation is noted from the top to the bottom of the image. (B) On higher power, there is immature malignant "lace-like" osteoid between cells.

Pathologic findings

The right adnexa measured 25.0 × 18.0 × 8.0 cm in size. Grossly, it was gray-brown and elastic. Upon dissecting the tumor, it was multiloculated with mucinous content. On serial cut, a red-brown and elastic mural nodule was noted, measuring 5.0 × 4.5 × 4.0 cm in size. Focal calcification or ossification was also noted in the mural nodule and septa. Fimbria-like tissue was identified.

Microscopically, the cystic tumor showed an ovarian mucinous cystadenoma with focal epithelial proliferation accounting for about 5% of the lining epithelium. There was no obvious cytologic atypia.

The mural nodule was composed of atypical spindles to epithelioid stromal cells with several osteoclast-like giant cells in a hemorrhagic background. Mitosis was focally brisk. Florid osteoid formation, including lace-like osteoid, was observed. Bony trabeculae and focal chondroid differentiation were also noted. There were also some small foci showing a similar histological picture on the surface of elsewhere septa. There was no anaplastic carcinoma or teratomatous component in the histological sections.

Immunohistochemically, the atypical spindle cells were positive for vimentin, whereas the giant cells were positive for CD68. Scattered CK-immunoreactive cells were also identified.

The histology of the mural nodule itself was consistent with a giant cell-rich osteosarcoma. The possibility of a sarcoma-like nodule was less likely, due to the presence of a lace-like osteoid and florid ossification. The overall picture suggested a mucinous cystadenoma with a malignant osteosarcomatous mural nodule.

The fimbria of the fallopian tube was identified and exhibited minimal histological change.

Representative images of the tumor are presented in Fig. 1.

Discussion

Mucinous cystadenoma of the ovary is the benign form of mucin-containing epithelial ovarian tumors. The peak incidence is at 30–50 years old. The tumor comprises 80% of mucinous ovarian tumors and 20–25% of all benign ovarian tumors. The tumors are lined by columnar epithelium, which are typically similar to endocervical epithelium, although an intestinal type may occasionally be seen. Mucinous cystadenomas are benign and have excellent prognosis.

Some cases of mural nodules found in ovarian mucinous tumors have been reported. The histologic types of mural nodules include sarcoma-like mural nodules [1], carcinosarcoma-like mural nodules [4], true sarcoma, and anaplastic carcinoma. Sarcoma-like mural nodules may be reactive in nature and are composed of a very cellular and heterogenous population of fibroblasts and inflammatory cells. Mitosis may be conspicuous in the fibrohistiocytic cells. However, most mural nodules are malignant according to the reports in the literature.

It is important to distinguish sarcoma-like mural nodules from sarcoma or anaplastic carcinoma due to the difference in their prognoses. The sarcoma-like mural nodules are presumably reactive, and the impact on prognosis is negative [1]. However, most patients with mural nodules of anaplastic carcinoma present with a rapid progression of their disease and have poorer outcomes [5–7].

Reviewing the literature, there are only two cases of an osteosarcoma occurring as a mural nodule in an ovarian mucinous neoplasm [3]. In our case, there was a sharp demarcation between the benign mucinous cystadenoma and the mural nodule. The nuclear atypia of stromal cells, including atypical mitotic activity, confirmed the diagnosis of malignancy. Additionally, osteoclast-like giant cells and osteoid formation indicate the characteristics of osteosarcoma.

Osteosarcoma as a primary ovarian neoplasm is rarely reported, either as a pure osteosarcoma or arising in a teratoma [8]. The pathogenesis of osteosarcoma is not well known. It is believed to develop from the bone component of a teratoma, malignant mixed mesodermal tumor, or primitive ovarian stromal cells/metaplastic stromal cells. There are also some reports of metastatic osteosarcomas of the ovary [9].

The cases of primary ovarian osteosarcoma are often diagnosed at an older age with advanced stages and poor prognosis. The only two cases with FIGO stage I disease received adjuvant chemotherapy after operation, and showed no evidence of recurrence with follow-up at 3 years and at 16 months [8,10].

The two cases previously reported as mural nodules of an osteosarcoma arising from an ovarian mucinous tumor occurred at younger ages (18 and 34 years old), and both patients were at FIGO stage I. They did not receive adjuvant chemotherapy after surgery and had no recurrence of the disease at 12- and 18-month follow-up [3], which is similar to our present case at 16-months follow-up.

Although ovarian osteosarcomas have a high (>60%) case fatality rate, the survival rate strongly depends on the advanced stage at which most patients present. The outcome may be favorable either in stage I primary ovarian osteosarcoma or in osteosarcoma as a mural nodule in ovarian mucinous neoplasms. However, there is no consensus regarding the treatment strategy for osteosarcomatous mural nodules in mucinous tumors because of the rarity of the disease. More case studies need to be performed before the pathogenesis of this lesion can be fully sustained.

Funding

None.

Declaration of competing interest

None.

Acknowledgements

None.

References

[1] Prat J, Scully RE. Ovarian mucinous tumors with sarcoma-like mural nodules: a report of seven cases. Cancer 1979;44(4):1332–44.
[2] Chen TY, Chen BF, Chen CK, Wang KL, Yang YC, Wang TY. Ovarian borderline mucinous tumor with stromal microinvasion and hemangiomatous mural nodules. Int J Gynecol Pathol 2010;29(3):239–42.
[3] McFarland M, Dina R, Fisher C, McCluggage WG. Osteosarcoma as malignant mural nodule in ovarian mucinous neoplasms of intestinal type: report of 2 cases. Int J Gynecol Pathol 2015;34(4):369–73.
[4] Chang WC, Sheu BC, Lin MC, Chow SN, Huang SC. Carcinosarcoma-like mural nodule in intestinal-type mucinous ovarian of borderline malignancy: a case report. Int J Gynecol Cancer 2005;15(3):549–53.
[5] Baergen RN, Rutgers JL. Mural nodules in common epithelial tumors of the ovary. Int J Gynecol Pathol 1994;13(1):62–72.
[6] Chan YF, Ho HC, Yau SM, Ma L. Ovarian mucinous tumor with mural nodules of anaplastic carcinoma. Gynecol Oncol 1989;35(1):112–9.
[7] Prat J, Young RH, Scully RE. Ovarian mucinous tumors with foci of anaplastic carcinoma. Cancer 1982;50(2):300–4.
[8] Hines JF, Compton DM, Stacy CC, Potter ME. Pure primary osteosarcoma of the ovary presenting as an extensively calcified adnexal mass: a case report and review of the literature. Gynecol Oncol 1990;39(3):259–63.
[9] Eltabbakh GH, Belinson JL, Biscotti CV. Osteosarcoma metastatic to the ovary: a case report and review of the literature. Int J Gynecol Pathol 1997;16(1):76–8.
[10] Ngwalle KE, Hirakawa T, Tsuneyoshi M, Enjoji M. Osteosarcoma arising in a benign dermoid cyst of the ovary. Gynecol Oncol 1990;37(1):143–7.