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Uncommon malignancy arising within mature ovarian teratoma: Case report and literature review

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ABSTRACT

Background. Clear cell carcinoma of the ovary developing in the background of an ovarian teratoma represents an extremely rare entity of the gynecologic tract. Giving the rarity of this entity, the histologic challenges, prognosis and the adequate treatment is still uncertain.

Methods. We herein present a case of clear cell carcinoma coexisting with a teratoma, accompanied by a scrutinous literature review. The authors have reviewed all case reports published in the literature (1978-2021), adding our new case, in order to enrich current literature data.

Results. A thorough search of the PubMed/MEDLINE and Google Scholar databases has revealed 7 cases of clear cell carcinoma of the ovary developing in association with a teratoma. Out of the 7 cases, 4 were part of a more complex tumor, including other malignancies (e.g. angiosarcoma, endometrioid carcinoma). The mean size of the tumors was 16 cm and most patients died of disease or suffered multiple recurrences. Additionally, we report a case of a 54-year-old patient who presented with an ovarian tumor and which upon microscopic examination featured a cystic structure lined by squamous keratinizing epithelium and sebaceous glands. Inside the wall of the cyst, a small nodule made out of tubules and papillae lined by clear cells, which invaded the surrounding stroma was identified. Four months later, the patient developed a small nodule in the liver and she has subsequently received adjuvant chemotherapy.

Conclusion. Clear cell carcinoma of the ovary developing in association with a teratoma is an extremely rare entity, that requires thorough sampling of the cyst and which is usually associated with a relatively poor prognosis.

Keywords: clear cell carcinoma, ovarian teratoma, malignancy within teratoma

INTRODUCTION

Malignant tumors developing in association with mature or immature tumors represent rare tumors, comprising less than 2,7% of all teratomas [1,2]. Squamous cell carcinoma is the most common epithelial malignant neoplasm developing within a teratoma, representing 80% of all malignant mature cystic teratomas [1]. In comparison, clear cell carcinoma of the ovary developing in the background of a mature teratoma is an exceptional situation, with only 7 cases reported in the scientific literature in the timeframe 1978-2021. Even though the scientific data regarding this particular entity are scarce, a recent study demonstrated that both

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tumors harbor ARID1A deficiency and also PIK3CA mutation [3].

MATERIAL AND METHOD

For the purpose of analyzing the particularities of ovarian clear cell carcinoma which coexists with teratoma, we have performed a literature search on the several MESH terms: "ovarian" "clear cell carcinoma", "teratoma", "endometriosis", "mixed germ cell tumor" and "mixed mesodermal tumor". The entire PubMed®/MEDLINE and Google Scholar databases were analyzed and all relevant scientific papers were reviewed and compared to the case presented below. All the cases were sorted by year and the following information was retrieved: age of the patient, size of tumor, presenting symptoms, surgical and oncological treatment and also the prognosis. PubMed®/MEDLINE data search for case reports revealed 7 cases published in the last 43 years.

RESULTS

The performed literature review revealed 7 cases of ovarian clear cell carcinoma coexisting with teratoma, 4 out of which also associated other tumors (e.g. angiosarcoma, endometrioid carcinoma). Mean size of the tumors was 16,18 cm and the most common symptoms were abdominal pain or distension. 83% of all cases underwent total abdominal hysterectomy (TAH), bilateral salpingo-oophorectomy (BSO) with adjuvant chemotherapy. Out of the scientific papers that reported survival outcome data, 50% died of disease (DOD) in less than 17 months, while the other 50% either featured multiple recurrences, or were disease free after 8 months of follow-up.

CASE PRESENTATION

We report a 54-year-old patient, which presented to the hospital for abdominal discomfort, that increased in intensity in the last days. A CT examination was performed, which revealed a large tumor of the right ovary, with a cystic structure. An intraoperative examination has been performed and the preliminary diagnosis was mature teratoma. Upon gross examination, the tumor had a cystic structure with an intact capsule, that featured a thickening of the cyst wall with irregular internal surface and firm consistency. The whole tumor measured 14x12x12 cm, and the thickened area measured 1.7x1.5x0.9 cm. A thorough sampling of the wall cyst and of the area thickened has been performed. Upon microscopic examination, the cystic structure was lined by a squamous keratinizing epithelium, which included with hair follicles and sebaceous glands, representing a mature teratoma. Sampling from the thickened wall cyst revealed a secondary proliferation constituted out of tubule-cystic and papillary structures lined by cuboidal cells with pale or clear cytoplasm and vesicular nuclei and prominent nucleoli (Figure 1). Some of the papillae were lined by cells with a hobnail aspect and had a hyalinized fibrovascular core (Figure 2). Several areas of stromal invasion were noticed, which featured cholesterol clefts, areas of necrosis and psammoma bodies (Figure 3). The described aspects were diagnostic for clear cell carcinoma of the ovary. Immunohistochemical test revealed that the malignant proliferation was positive for PAX8 and Napsin A, and had a proliferative index of 15%. The case has been discussed in the multidisciplinary board meeting and a complex course of adjuvant chemotherapy has been recommended, because the patient had developed liver metastases in less than 2 months from the diagnosis. She is currently closely monitored with imagistic analyses and oncological consults at each 6 months.

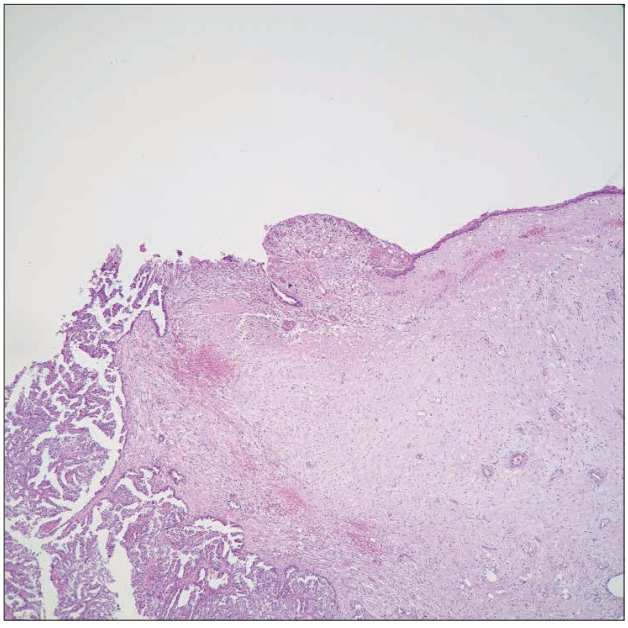

FIGURE 1. Transition area between squamous epithelium of teratoma and papillary proliferation of clear cell carcinoma (H.E., 40x)

DISCUSSIONS

In the recent scientific literature there have been many malignancies reported, which are known to arise in association with a mature teratoma, with the most common entity being represented by squamous cell carcinoma. Additionally, other subtypes reported in the literature are: sebaceous carcinoma, thyroid papillary carcinoma, mucinous adenocarcinoma, mucoepidermoid carcinoma, urothelial car-

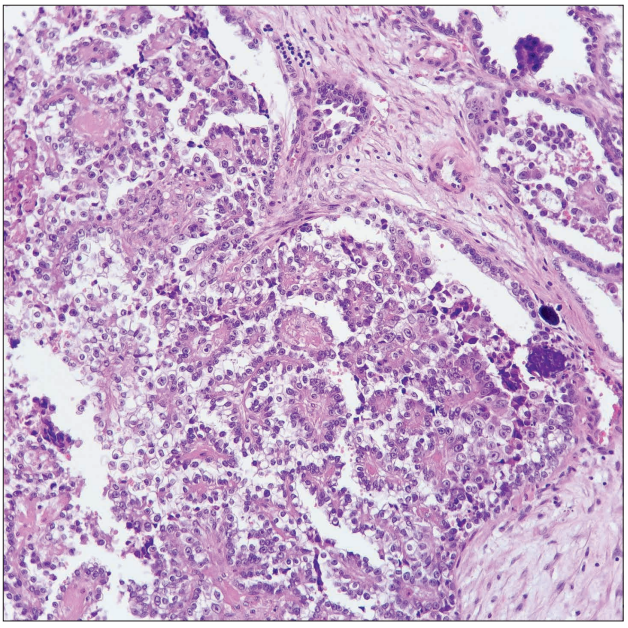

FIGURE 2. High magnification view of hyalinized fibro-vascular cores lined by clear cells with hobnail cells and focal calcifications (H.E., 200x)

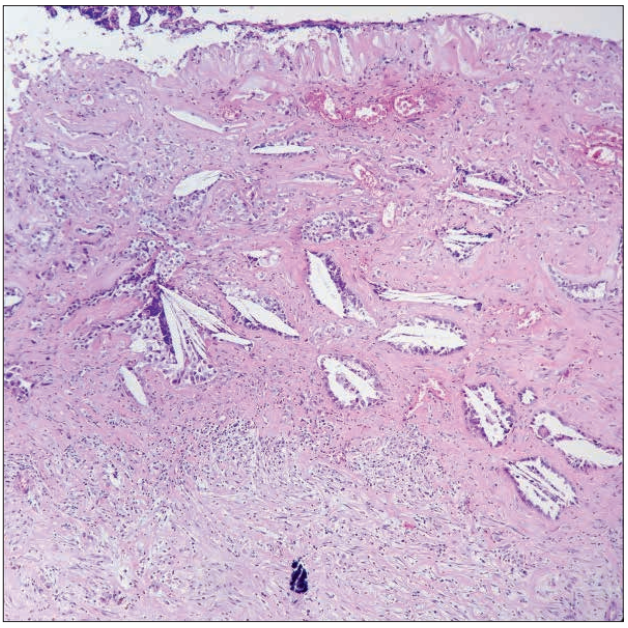

FIGURE 3. Medium power view showing invasive areas with cholesterol clefts lined by polygonal cells with clear cytoplasm and prominent nucleoli (H.E., 100x)

cinoma, carcinoid, carcinosarcoma, oligodendroglioma, neuroblastoma, meningioma, melanoma, undifferentiated pleomorphic sarcoma, leiomyosarcoma, rhabdomyosarcoma, plasmablastic lymphoma, diffuse large B cell lymphoma, follicular lymphoma [4–20]. Cases of teratoma localized in non-ovarian sites have also been reported to be associated with the following tumors: nephroblastoma (retroperitoneum), apocrine carcinoma (mediastinum), signet ring cell carcinoma (sacrococcygeal), salivary gland carcinoma (mediastinum), neuroendocrine carcinoma (retroperitoneum), papillary renal cell carcinoma (retroperitoneum) [21-26]. Additionally, Pires et al. have also reported the coexistence of an ovarian teratoma with a uterine adenocarcinoma [27]. One particular scientific article reported the development of a rhabdomyosarcoma inside an ovarian carcinoma, in a patient that had contralateral serous carcinoma [17]. Cagino et al. have reported a case of multiple malignancies arising in a monodermal teratoma, which encompassed papillary thyroid carcinoma and ovarian carcinoid, arising in the background of a struma ovarii [28]. Savitchi et al. have also reported a squamous carcinoma coexisting with a pleomorphic carcinoma, which developed inside a mature cystic teratoma [29]. Myeloid sarcoma has also been reported as a neoplasm arising in a mixed germ cell tumor of a mediastinum, which has a teratomatous component [30].

Notably is the development of several reports of N-methyl-D-aspartate receptor encephalitis in patients with ovarian teratoma [31-33] and also the development of paraneoplastic neuromyelitis [34]. Of significant clinical importance is the observation made by Belge et al., who noticed that microRNA-375-3p serum levels can be used as a reliable biomarker of teratoma [35].

Ovarian teratoma can rarely be part of a mixed germ cell tumor, case in which it can be associated with an embryonal carcinoma [36,37]. Similarly to ovarian teratoma, testicular teratoma can also be part of a mixed germ cell tumor, and can frequently associate a seminoma, an embryonal carcinoma, a choriocarcinoma or a yolk sac tumor component [38]. Similarly, mixed germ cell tumors with a teratomatous component can arise also in the third ventricle and can even cause leptomeningeal spread [39].

Cases of sebaceous carcinoma and squamous cell carcinoma arising in an ovarian teratoma have been demonstrated to harbor mismatch repair deficiencies [40].

The scientific literature has revealed only 7 published cases in the time frame 1978-2021 of clear cell carcinoma coexisting with a teratoma. Out of these 7 cases, only three of them were pure clear cell carcinomas arising from a teratoma (two mature, one immature), the other were part of a more complex tumor and featured tumors such as: one angiosarcoma, one malignant mesodermal tumor, one malignant mixed germ cell tumor (yolk sac tumor, embryonal carcinoma and mature teratoma) and one who also featured other multiple malignancies (endometrioid adenocarcinoma, yolk sac tumor, squamous cell carcinoma and neuroectodermal tumor with rhabdomyosarcomatous differentiation) [3,41-46]. No differences in the overall surviv-

TABLE 1

Article author	Age	Size	Symptoms	Stage	Treatment	Prognosis
Cooper et al. (1978) [41]	53	16X10X10 cm	Nocturia, low back pain	NA	NA	NA
	34	14 cm	Abdominal pain	pT1aN1M0	Right salpingo-oophorectomy, lymph node dissection, omentectomy, chemotherapy	Multiple recurrences after 5 and 17 months. Alive at 40 months after diagnosis.
Ohishi et al. (2007) [42]	59	16x12x4,5 cm	Abdominal mass	NA	TAH, BSO, omentectomy, chemotherapy (paclitaxel, platinum)	NA
	55	5,1x5x3,4 cm	Lower abdomen discomfort		TAH, BSO, omentectomy, right infundibulopelvic ligament resection and bilateral paracolic sulci peritoneum biopsies, Chemotherapy (bleomycin, etoposide, cisplatin)	NA
Takahashi et al. (2012) [43]	49	16x10x9,5 cm	Genital bleeding	pT1aN0Mx	TAH, BSO, omentectomy, lymph node dissection, chemotherapy (taxol, carboplatin,	DFS 8 months
	71	30 cm	Abdominal distension	pT2aNxM1	TAH, BSO, chemotherapy	DOD 17 months after diagnosis
Yu et al. (2014) [44]	47	NA	Abdominal distension	pT1c	TAH, BSO, lymph node dissection, chemotherapy	DOD 9 months after the surgery

al of patients with only clear cell carcinoma and teratoma and those with clear cell carcinoma, teratoma and other malignancies.

The first case described had similarities with the case presented above, namely both featured areas of endometriosis [41]. This finding can support the presumption that both tumors develop from the Mullerian/ paramesonephric tissues[41]. Kihara et al. have analyzed the molecular similarities between the clear cell carcinoma and the immature teratoma and noticed that both tumors featured ARID1A deficiency and also PIK3CA mutation [3].

A mesenchymal component with different morphologies (angiosarcoma, rhabdomyosarcomatous and cartilaginous) has been noted in three out of seven cases [41-43].

CONCLUSIONS

We presented an extremely rare case of an ovarian clear cell carcinoma developing in association with a mature teratoma, in the background of endometriosis. These rare malignancies are known to have an adverse prognosis with a short disease-free survival and a high mortality rate. This case can emphasize the need to scrutinous analyze the slides when dealing with a teratoma, or with a tumor of Mullerian origin. A malignant component, when present, should always be reported and the pathologists need to be aware of such rare associations.

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