

Case Report

# “Tumor in tumor”: A Rare Carcinoma Arising in Benign Cystic Teratoma of Ovary

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## Abstract

**Introduction:** Mature cystic teratoma is the most common germ cell tumor of the ovary. Case reports of malignant transformation in these benign tumors have been reported. The most common malignancies arising in the background of mature teratomas are squamous cell carcinoma and adenocarcinoma. Skin appendageal tumors also have been reported of which most are sebaceous carcinomas. .

**Case Presentation:** A 54-yr-old lady presented with abdominal pain and abdominal mass. Radiological investigations revealed a complex cystic mass in the left ovary. A total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. Histopathological examination revealed sebaceous carcinoma in-situ arising in a mature cystic teratoma of ovary.

**Conclusion:** Sebaceous carcinoma in-situ is extremely rare occurrence in mature teratoma of ovary. However it should be keenly looked for as other carcinomas so that the patient can be kept on regular follow up. This is the first report of such a rare occurrence.

**Keywords:** teratoma; sebaceous carcinoma; sebaceous carcinoma-in-situ; ovary; malignancy

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**Consent:** Consent was taken from the patient for publication of this case report.

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## Introduction

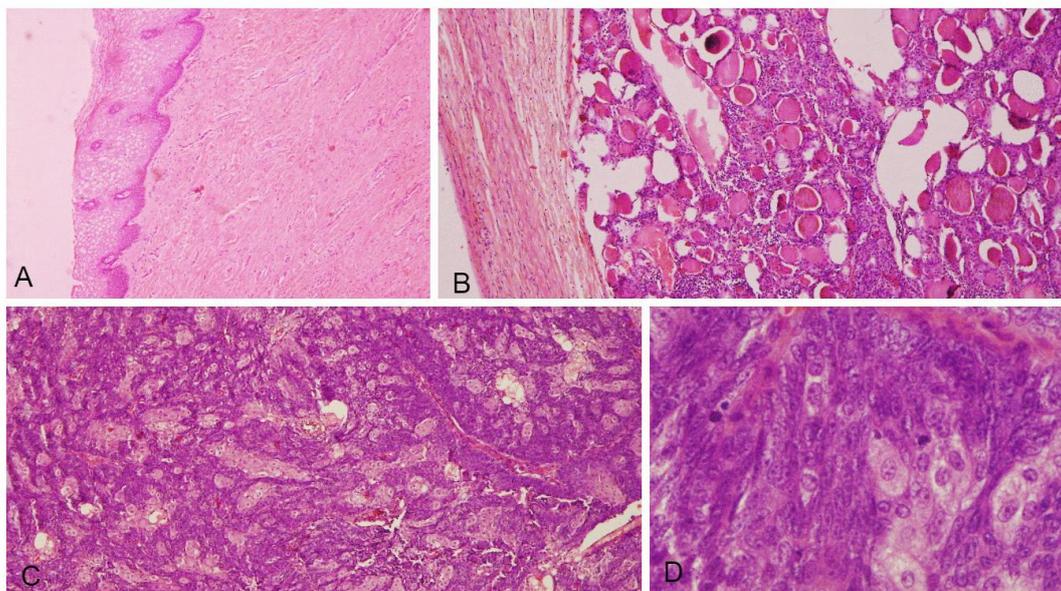
Mature cystic teratoma of the ovary is a benign tumor. It is classified under germ cell tumors and histologically it is representative of one or more germ layers<sup>1</sup>. There have been reports of malignant change arising in the mature tissues seen in the teratomas, the most common malignancy being squamous cell carcinoma<sup>2</sup>. Skin adnexal neoplasms are of very low incidence. Sebaceous neoplasms are extremely rare and all of the reported cases are of sebaceous carcinoma. We present a case of sebaceous carcinoma in situ arising in mature cystic teratoma of ovary along with a brief review of literature.

## Case Presentation

A 54-year-old woman was admitted to our hospital with complaints of abdominal distention and lower abdominal pain of 6 months duration. Abdominal examination revealed a palpable left adnexal mass. There was no peripheral or intra-abdominal lymphadenopathy. The serum CA-125 level was mildly elevated. Computed tomography showed a heterogenic tumor of the right ovary. A total abdominal hysterectomy with bilateral salpingo-oophorectomy and omentectomy was performed.

Grossly, the left ovary was enlarged and measured 13x8 cm in maximum dimension. Externally the ovary was irregular, however the capsule was intact. On cut section, the entire ovary was replaced by a predominantly cystic cavity filled with hair and cheesy material. The inner wall of the cyst was smooth. An intramural solid component measuring 2x1cms was identified which was processed in entirety.

Microscopically, the cyst showed mature tissue representing epidermis (Figure A), hair follicles, sweat glands, salivary gland, respiratory epithelium, transitional epithelium, lymphoid aggregates and thyroid parenchyma (Figure B). In addition, there was a well circumscribed sebaceous component comprising of mature lobules of sebaceous gland. A focal area within the sebaceous lobule showed presence of basaloid cells with scant amount of cytoplasm and hyperchromatic nuclei (Figure C). These basaloid cells were not surrounded by a peritumoral myxoid stroma and tongues of basaloid cells infiltrating into the surrounding stroma were not seen. These cells were intimately intermixed with slightly larger cells having a finely vacuolated cytoplasm and vesicular nucleus with clumped chromatin and prominent nuclei. These cells were smaller in size than the fully differentiated sebaceous cells and showed more hyperchromasia. There was brisk mitotic activity with mitotic figures ranging from 2-4 per high power field (Figure D). Foci of necrosis were also seen. This entire focus was surrounded by mature sebaceous gland which in itself was quite well circumscribed and was separated from the other tissues by a thin fibrous band (Figure C). However, even after extensive serial sectioning and examination of more tissue from the ovary no other such area could be identified. Lymphovascular emboli were absent. There was no focus of invasion into the adjoining components or the capsule of the ovary. Hence, a diagnosis of mature cystic teratoma with sebaceous carcinoma in situ was proffered. The right tube and ovary was histologically unremarkable. The uterus showed an intramural leiomyoma. The omentum was free of tumor.



**Fig.1** (A) Epidermis along with hair follicle, lymphoid aggregates and thyroid parenchyma. (B) Lobules of mature sebaceous glands adjacent to sweat ducts and thyroid parenchyma. (C) Sheets of basaloid cells admixed with the mature sebaceous gland. (D) Brisk mitotic activity is seen in the basaloid cells.

## Discussion

Mature cystic teratomas account for 10 to 20% of ovarian neoplasms. They are also the most common ovarian germ cell tumor. They are bilateral in 8-14% of cases [1-3].

Cystic teratomas of the ovary are diagnosed most frequently during the reproductive years. The peak incidence in most series is age 20-40 years [1,3].

It is considered to be a benign tumor however malignant transformation has been reported especially in the postmenopausal age group [4]. The most common malignancy reported is the squamous cell carcinoma. The others are adenosquamous carcinoma, adenocarcinoma, melanoma and basal cell carcinoma [5-9].

There are very few case reports of sebaceous carcinoma and none for sebaceous carcinoma-in-situ.

Sebaceous carcinomas are infiltrating neoplasms. They are believed to arise from mature sebaceous cells which undergo malignant change in ovarian teratomas [11]. Some authors propose that these neoplasms may arise from pluripotent stem cells.

Few authors have studied the immunohistochemical profile of the sebaceous carcinomas arising in teratomas. There is overexpression of hormonal receptors and mutant p53. Low ki67 index has been reported to have a favourable prognosis [11]. However none of these markers have any effect on the overall prognosis or clinical outcome and hence are not mandatory for the diagnosis.

Sebaceous carcinomas have to be differentiated from basal cell carcinomas and sebaceomas (sebaceous epitheliomas). Basal cell carcinomas have infiltrating borders, peritumoral myxoid stroma and peripheral palisading of cells. Sebaceomas are well circumscribed nodules or irregularly shaped cell masses in which more than half of the cells are undifferentiated basaloid cells but in which there

are significant aggregates of mature sebaceous cells and of transitional cells. Though mitotic activity may be present, sebaceous epitheliomas lack nuclear atypia and invasive, asymmetric growth patterns, which are hallmarks of sebaceous carcinoma.

Sebaceous carcinoma-in-situ shows morphological features similar to sebaceous carcinoma. However, in addition they are surrounded by mature and well differentiated sebaceous glands and are limited to the sebaceous lobule without infiltration into the surrounding structures. There is no peritumoral myxoid stroma or peripheral palisading, unlike those to basal cell carcinomas. The mature sebaceous glands predominate over the basaloid cells in sebaceous epitheliomas differentiating them from sebaceous carcinomas and sebaceous carcinoma-in-situ.

The prognosis of any malignancy arising in teratoma depends on the stage, vascular and capsular invasion. Sebaceous carcinomas reported till date have not shown any recurrence or metastasis suggesting these malignancies have a favorable prognosis [12-14]. Our case was stage I and underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy and omentectomy. The patient is doing well on follow up and has no evidence of recurrence or metastasis.

To the best of our knowledge this is the first description of a case of sebaceous carcinoma-in-situ. Although very rare, it should be included in the differential diagnosis of skin adnexal tumors arising in teratoma of ovary.

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