Sebaceous adenoma arising in mature cystic teratoma of the ovary. Case report

Kristýna Němejcová¹, Pavel Dundr¹, Jana Rosmusová¹, Inna Tučková²

¹Department of Pathology, First Faculty of Medicine and General University Hospital, Charles University in Prague, Czech Republic
²Department of Pathology, Military University Hospital in Prague, Czech Republic

SUMMARY
We report the case of a 44-year-old female with sebaceous adenoma arising in mature cystic teratoma of the ovary. The patient had a tumor in the left ovary; 125 x 90 x 70 mm. Microscopically, the tumor consisted of structures typical of dermoid cysts. However, large areas of sebaceous proliferation were found. These areas were comprised of sebaceous nodules with features similar to a sebaceous adenoma of the skin. Immunohistochemically, the tumor showed "wild-type" expression of p53 and low proliferative activity (Ki-67 index < 5%). To verify the possibility of Muir-Torre syndrome we performed immunohistochemical examination of DNA mismatch repair proteins expression. However, all four proteins examined (MSH2, MSH6, MLH1, PMS2) were positive. Sebaceous adenoma arising in mature teratoma of the ovary is rare. To the best of our knowledge, only six cases have been reported in the literature to date.

Keywords: sebaceous adenoma – mature cystic teratoma – ovary

RESULTS
Grossly, the left adnexal tumor measured 125 x 90 x 70 mm. The dissection of the tumor revealed a cavity filled with fatty material similar to normal sebum, and hair surrounded by a firm capsule of varying thickness. The inner lining of the cyst was predominantly smooth, with only one yellowish tumorous mass measuring 40 x 30 x 10-15 mm.

Microscopically, the majority of the tumor was composed of mature cystic teratoma structures, including skin and skin adnexa, and also contained respiratory-type epithelium. However, the whole yellowish tumorous mass showed an abnormal proliferation of sebaceous cells forming nodules with features similar to a sebaceous adenoma of the skin (Fig. 1). This part of the lesion had a lobular growth pattern and a pushing border with ad-
The histologic spectrum of sebaceous lesions and tumors encompasses sebaceous hyperplasia, sebaceous adenoma, sebaceoma and sebaceous carcinoma (10). Sebaceous adenoma has to be differentiated from sebaceous hyperplasia, in which the sebaceous lobules are increased in number, but comparing with sebaceous adenoma have only two layers of peripherally located basaloid or germinative cells (11). There can be some histological overlaps between sebaceous adenomas and sebaceomas. Sebaceomas are irregularly shaped nodular lesions comprising undifferentiated basaloid sebocytes in more than half of the tumour cell volume, and to a lesser extent small groups of sebaceous cells and transitional cells. Sebaceous adenomas and sebaceomas, in contrast to sebaceous carcinoma, lack nuclear atypia and invasive growth patterns. However, there may be substantial mitotic activity present in the basaloid regions in these benign tumors. Sebaceous carcinomas are cytologically and/or architecturally malignant tumors with sebocytic differentiation and the grading of these carcinomas is based on growth patterns rather than on their cytological features (10). Regarding immunohistochemical expression, analysis of p53 and Ki-67 may be helpful in differential diagnosis between benign and malignant sebaceous proliferations. One study has shown that sebaceous hyperplasia, sebaceous adenomas, and sebaceomas tended to show low levels of p53 and Ki-67 positivity, whereas sebaceous carcinomas tended to show higher levels of nuclear p53 expression (50% versus 11%) and Ki-67 positivity (30% versus 10%) compared to the adenomas (12).

The outcome of sebaceous adenomas arising in ovarian teratoma is favorable; all patients were well and disease-free for peri-jacent ovarian stroma. The lobules were composed of two cell types, cuboidal peripheral germinative cells and central mature sebaceous cells (Fig. 2,3). Immunohistochemical analysis of p53 exhibited weak nuclear positivity of scattered cells and moderate nuclear positivity in sporadic cells, in keeping with “wild-type” expression. The Ki-67 proliferation index was low, less than 5% of all tumor cells revealed nuclear positivity, with only some sporadic foci of “hot-spots”, where positivity reached 25% of tumor cells. Immunohistochemistry of MMR proteins showed nuclear positivity with antibodies against all four proteins examined (MSH2, MSH6, MLH1, PMS2) (Fig. 4).

DISCUSSION

Mature cystic teratoma is the most common type of ovarian teratoma and the most common type of ovarian germ cell neoplasm. It comprises approximately 20% of all ovarian neoplasms (5). Tumors with sebaceous differentiation arising in mature cystic teratomas are rare, although sebaceous glands are almost always components of mature cystic teratomas. These tumors include sebaceous adenoma, basal cell carcinoma with sebaceous differentiation, and sebaceous carcinoma. There have only been six prior reports of sebaceous adenoma, nine reports of sebaceous carcinoma, and two reports of basal cell carcinoma with sebaceous differentiation arising in mature cystic teratoma of the ovary (1-4,6-9). The sebaceous adenomas were in all cases composed of nodules or lobules of proliferating sebaceous cells showing various degrees of maturity, with mature cells predominating.

![Fig. 1. Sebaceous adenoma surrounded by a connective tissue capsule of varying thickness (H&E, original magnification 20x).](image1)

![Fig. 2. Sebaceous adenoma composed of nodules of sebaceous cells (H&E, original magnification 100x).](image2)

![Fig. 3. Two cell types, peripheral germinative cells and central mature sebaceous cells, are present (H&E, original magnification 400x).](image3)

![Fig. 4. Immunohistochemical examination of MMR proteins showed nuclear positivity with antibodies against all four proteins examined. A: MLH1. B: PMS2 showing nuclear positivity (original magnification 200x).](image4)
In conclusion, we report a case of sebaceous adenoma arising in mature cystic teratoma of the ovary. To the best of our knowledge, only six cases of such a tumor have been reported in the literature to date.

ACKNOWLEDGEMENTS

This work was supported by Charles University in Prague (Project PRVOUK-P27/LF1/1, UNCE 204024, Ministry of Health, Czech Republic - conceptual development of research organisation 64 165, General University Hospital in Prague, Czech Republic, and by OPPK (Research Laboratory of Tumor Diseases, CZ.2.16/3.1.00/24509).

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest regarding the publication of this paper.

REFERENCES