CASE REPORT

Rare association of epidermoid cyst of ovary with mucinous cystadenoma- A Case Report

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Abstract

Epidermoid cyst is a rare benign neoplasm of the ovary, lined by stratified squamous epithelium, lacking skin, adnexal structures, and other teratomatous elements. On the other hand, mucinous cystadenoma is one of the most common benign ovarian neoplasm that microscopically shows cystic areas lined by tall columnar mucinous epithelium. Although its coexistence with other tumours, such as mature cystic teratoma, squamous cell carcinoma, clear cell adenocarcinoma, Brenner's tumour, serous cystadenoma, etc., have been reported, a combination of benign epidermoid cyst and mucinous cystadenoma has rarely been documented in literature. We report a case of coexistence of epidermoid cyst and mucinous cystadenoma in an ovarian cyst.

Keywords: Epidermoid cyst, Mucinous cystadenoma.

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Introduction

Epidermoid cyst (EC) of ovary is very rare and only about 40 cases of ovarian epidermoid cyst have been reported to date.[1] EC is lined by benign stratified squamous epithelium and the lumen is filled with keratin flakes. It is distinguished from mature cystic teratoma by the absence of skin adnexal structures and other teratomatous components.[2] The histogenesis of EC in the ovary is still debatable.[1] It probably arises from the epidermis which gets misplaced after injury or due to developmental anomaly of follicular infundibulum of the skin. EC of the ovary may be a part of mature cystic teratoma in which the skin's adnexal structures are either missed due to lack of meticulous sampling or they may be entirely absent.[3]

Mucinous cystadenoma (MC) is a very common benign neoplasm of the ovary[4] and arises from the ovarian surface epithelium. Mucinous neoplasms usually occur in women in the third and fourth decades of life.[5] MC rarely affects children and adolescents.[6] It is usually lined by intestinal type epithelium with picket fence appearance, goblet cells, Paneth cells, and endocrine cells.

Co-existence of epidermoid cyst with mucinous cystadenoma of the ovary is exceptionally rare and is infrequently been reported in literature.[5] Clinical importance of this co-existence is not significant because of the entirely benign nature of both the entities; however, it is of academic interest to know that both entities can co-exist.

Case Report

A 14-year-old girl presented at Indus hospital And Health Network, Karachi on 6th August 2021 with the complaint of fever, abdominal pain, nausea, and loss of appetite for 10 days, and a history of irregular menstruation. Computed Tomography (CT) scan of the abdomen and pelvis showed a circumscribed fluid containing cystic lesion arising from the right hemipelvis with dimensions of 17.0 x 16.0 x 9.7cm. Probable origin of this lesion was the right adnexa. The lesion appeared well encapsulated without evidence of frank infiltration. The lesion was causing compression of both the ureters. Overall features favoured benign looking neoplastic lesion. Beta HCG and Alpha fetoprotein levels were within normal limits. CA-125 level was 38.51 IU/ml. The patient underwent exploratory laparotomy at Indus Hospital and Health

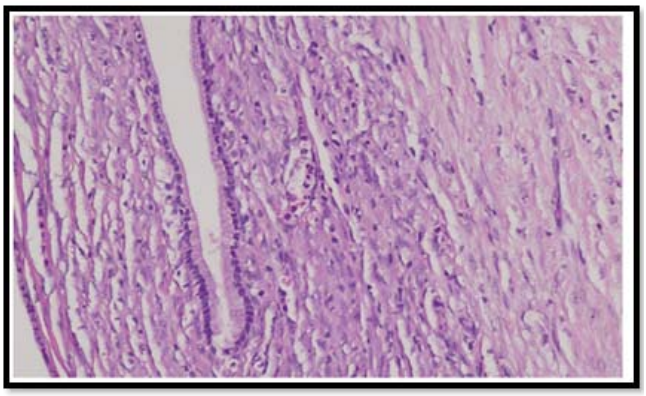

Figure-1 : H & E showing mucinous cystadenoma lined by tall columnar mucinous epithelium.

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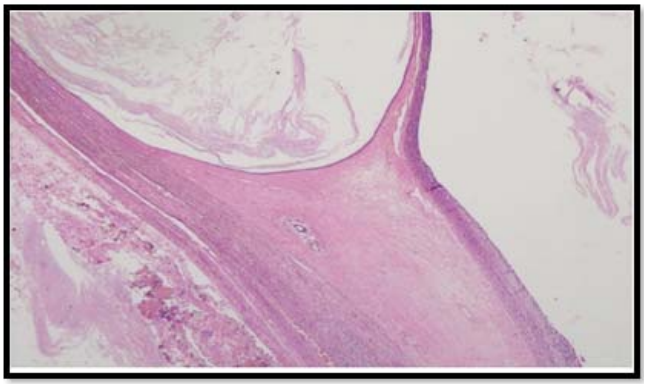

Figure-2: H & E showing both Epidermoid cyst (Left) and mucinous cystadenoma (Right).

Network, Karachi. Per-operatively a huge cystic mass was identified attached to the right fallopian tube. The cyst was excised and sent for histopathological examination. Grossly, the cyst measured 20 x 14 x 11cm and weighed 1,990 grams. On sectioning, it was a multiloculated cyst filled with pale yellow keratinous material. Microscopic examination revealed a cyst wall with extensively denuded lining epithelium. The cyst wall was predominantly lined by histiocytes and inflammatory cells. Focally, the cyst was lined by tall columnar mucin-secreting epithelium (Figure 1). No evidence of architectural complexity or nuclear atypia was seen. The cyst wall also showed a small cystic area lined by keratinized squamous epithelium. Keratinous material was seen in the lumen. No sebaceous glands or eccrine glands were seen in the cyst wall. Both these lining epithelia were intermingled in the cystic areas (Figure 2). CK7 and CKLMW immunohistochemical stains were positive in tall columnar mucin-secreting epithelium, while CK 20 was negative. Focal separately lying area adjacent to the mucinous areas of the cyst wall showed small glands surrounded by muscle tissue. A diagnosis of epidermoid cyst with associated mucinous cystadenoma was made. Verbal consent from the patient was taken and the patient was not asked to come for a follow-up visit.

Discussion

Epidermoid cyst of the ovary has been reported in the past in association with stromal hyperplasia,[3] in combination with well-differentiated endometroid adenocarcinoma of ovary and with primary carcinoid tumour of ovary.[7] Young and Scully proposed that epidermoid cysts originate from epithelial cell nests in the same manner as those seen in Brenner tumour.[1] Fan et al thought that these cysts are not as rare as we think since most of them may be misdiagnosed as mature cystic teratoma and epidermoid cysts are a subtype of mature cystic teratoma.[8] Nogales and Silverberg thought that epidermoid cyst arise from metaplasia of coelomic surface epithelium of the ovary.[9] Khidmati et al suggested that epidermoid cyst has heterogeneous origin and it may develop from teratoma, Brenner tumour, and endometriosis. It has also been suggested that epidermoid cyst develops from squamous epithelium implanted in the ovary during some previous surgery.[10]

Coexistence of mucinous cystadenoma has been reported with mature cystic teratoma, serous cystadenoma, carcinoid tumour, clear cell adenocarcinoma, squamous cell carcinoma, Brenner's tumour, endometriosis cyst, granulosa cell tumour, and with signet ring cells. Among these, a combination of mature cystic teratoma and mucinous cystadenoma is more common,[4] and others have been reported in rare case reports. Mucinous cystadenoma alone or in combination with mature cystic teratoma may rupture and cause pseudomyxoma paritonei. Co-existence of mucinous cystadenoma with Brenner's tumour, endometriotic cyst, granulosa cell tumour and serous cystadenoma is reported to have good prognosis in comparison to its combination with squamous cell carcinoma or clear cell adenocarcinoma which showed poor prognosis.[4] Mucinous cystadenoma with carcinoid tumour has intermediate malignant potential while prognosis of signet ring cells with mucinous cystadenoma is not reported.[4] Coexistence of epidermoid cyst and mucinous cystadenoma has been reported in a 37-year-old pregnant woman by Terada et al.[5] It was proposed that both epidermoid cyst and mucinous cystadenoma arise from surface epithelial cells and represent the same neoplasm.[4]

Conclusion

Both epidermoid cyst and mucinous cystadenoma are benign conditions and the management of both conditions is the same. Clinical significance of this combination has not been established yet. However, it is important to report this combination for the sake of academic interest.

Consent: The mother of the patient provided a written consent for publishing her case for promotion of science.

Disclaimer: None.

Conflict of Interest: We declare that the person who signed the ethical review statement is also a co-author of the manuscript.

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