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Case Report

Primary ovarian squamous cell carcinoma arising from an epidermoid cyst of Ovary- A rare case report with review of literature

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ABSTRACT

Primary ovarian squamous cell carcinoma is a rare entity that usually arises from the malignant transformation of a mature cystic teratoma, endometriosis or Brenner's tumor. The denovo occurrence of primary squamous cell carcinoma of the ovary without prior lesion is a rare entity accounting for less than 1%. However, ovarian squamous cell carcinoma arising from an epidermoid cyst is an infrequent entity and only one case has been reported in literature. The prognosis of such cases is quite poor and there are no definite guidelines for treatment. Here we present a case of a 50-year-old female diagnosed with primary ovarian squamous cell carcinoma arising from an epidermoid cyst of the ovary who was treated with surgery followed by chemotherapy along with the review of literature.

1. Introduction

Squamous cell carcinoma of the ovary is an uncommon entity that may be primary or resulting from malignant transformation of a benign cystic teratoma, Brenner's tumor, or metastasis from other organs.[1–7] The denovo occurrence of ovarian squamous cell carcinoma is extremely rare accounting for 1% of ovarian cancer that arises from squamous metaplasia of surface epithelial lining.[1,2,7–9] Till now 42 cases of primary ovarian SCC have been reported.[2] Whereas primary ovarian squamous cell carcinoma arising from an epidermoid cyst is an infrequent scenario and as per our knowledge only one case has been reported in the literature.[5]

It affects women between 27 years to 90 years with an average age of incidence being 52.9 years.[1] There is no protocol for the management of these cases to date due to the rarity of presentation.[1–6] However, radical surgery and platinum-based chemotherapy are used in the treatment of these patients. Also, the prognosis of the cases is poor with a 5-year survival rate of 15%-52%.[1]

Here we report a rare primary ovarian squamous cell carcinoma arising from an epidermoid cyst in a 50-year-old female.

2. Case Report

A 50-year-old postmenopausal female patient presented to the hospital with complaints of abdominal pain and increased frequency of micturition for 10 days. An abdominal mass of approximately 12X8X10 cm was detected clinically which was confirmed by ultrasonography followed by Magnetic resonance imaging. Her vulva, vagina, and cervix appeared normal and the liquid-based cytology showed no abnormal cells. She had no history of any other malignancies. Her MRI revealed a large solid cystic lesion in the mid pelvi -abdominal region closely abutting bilateral adnexa with a solid component in the right superolateral; aspect of around 6X6X5cm with no lymph node enlargement. Her routine investigations were within

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normal limits. The tumor marker CA 125 of the patient was 38.4 U/ml. Then the patient was planned for staging laparotomy and the specimen was sent for histopathological examination.

We received a specimen of total abdominal hysterectomy with bilateral salpingo -oophorectomy with omentum and lymph nodes. On gross examination, a cyst was found attached to the right ovary measuring 16X12X8 cm along with attached solid areas measuring 9X6X5 cm. (Figure 1) On opening the cyst, cheesy white material oozed out. The cut surface of the solid area showed homogenous white areas along with hemorrhagic areas. Representative sections were taken and processed.

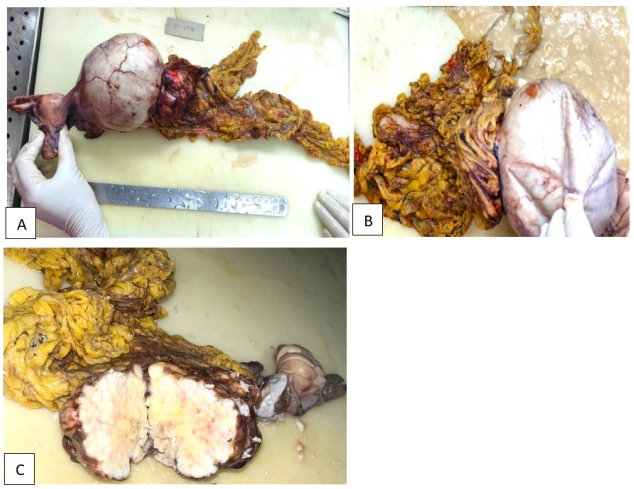

Figure 1: Gross: A cyst was found attached to the right ovary measuring 16X12X8 cm along with attached solid areas measuring 9X6X5 cm (**A**). On opening the cyst, cheesy white material oozed out (**B**); (**C**): The cut surface of the solid area showed homogenous white area

Microscopic examination of the cyst showed a keratinized stratified squamous epithelial lining with granular layer and the lumen was filled with keratin flakes. (Figure 2) The examination of solid areas revealed dysplastic keratinized stratified squamous epithelial lining showing hyperkeratosis and parakeratosis. The underlying stroma showed sheets of atypical squamous cells with increased nuclear-cytoplasmic ratio, pleomorphism, and hyperchromasia along with keratin pearl formation. (Figure 3) Sections from the contralateral ovary, cervix, and uterus were devoid of malignant cells.

Her chest X-ray, pelvic examination, and cervical cytology were normal without any malignancy. She had no symptoms suggestive of esophageal, bladder, lung, or anorectal pathology. So, considering all these, the final diagnosis of Primary Ovarian Squamous Cell Carcinoma arising from Epidermoid Cyst of the ovary was made and staged as FIGO Stage 1. The patient is now on treatment with 3 cycles of carboplatin 450 mg and paclitaxel 210 mg at 3-week intervals. Further, the patient will be reassessed after completion of treatment for the effect of chemotherapy and recurrence if any.

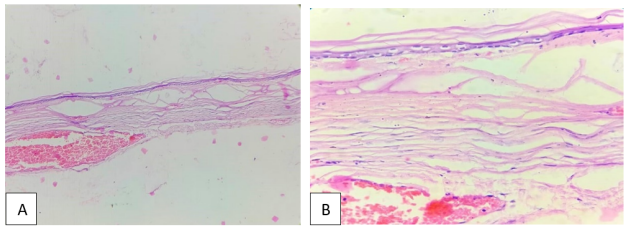

Figure 2: Microscopy: Sections show a cyst lined by keratinized stratified squamous epithelial lining with granular layer along with keratinous flakes (H&E-10X,40X)

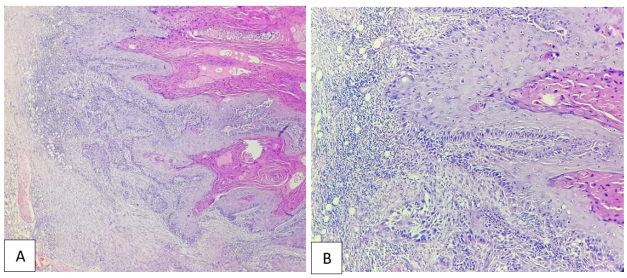

Figure 3: Microscopy: Section from solid area of right ovary shows dysplastic keratinized stratified squamous epithelial lining showing hyperkeratosis and parakeratosis. The underlying stroma showed sheets of atypical squamous cells along with keratin pearl formation (H&E- 10X, 40X)

3. Discussion

Epidermoid cyst is a benign lesion with keratinized stratified squamous epithelial lining with a lumen filled with keratin flakes. It is most common in skin. At the same time, the epidermoid cyst of the ovary is an extremely rare condition.[5,10,11] However, the diagnosis of a keratinous cyst should be made after a thorough examination of the whole specimen for any other components of ectodermal, mesodermal or endodermal derivative to rule out mature cystic teratoma.[5]

Ovarian squamous cell carcinomas are rare and aggressive tumors that account for less than 1% of ovarian tumors. 80% of such cases are derived from the mature teratoma, and less frequently from endometriosis or Brenner's tumors.[1–7] Primary ovarian squamous cell carcinoma can also occur due to metaplasia of surface epithelium or metastasis from uterine cervix, vagina, lung, esophagus or neck.[2,6] Black and Benitez et al. in 1964 described 42 cases of pure ovarian squamous cell carcinoma that had been reported in the literature.[2]

Primary ovarian SCC arising from an epidermoid cyst is extremely rare. Only one such case has been reported in the literature. Savita et al in 2017 reported the first case of primary ovarian SCC arising from an epidermoid cyst of the ovary in a 55-year-old postmenopausal female.[5] Sharma et al reported a case of a giant epidermoid cyst of the ovary, however, no malignant foci were identified.[10] Pins et al in 1996 reported 11 cases of pure ovarian squamous cell carcinoma among which none of the cases were associated with epidermoid cysts.[11]

The most common symptom is abdominal pain or abdominal mass. The diagnosis of primary ovarian SCC must be made after a thorough physical, radiological, and cytological examination of other possible sites of malignancies.[2,8,9,12] The Immunohistochemistry markers positive in primary SCC are CK34bE12 or CK5/6, p63.[2] About 36% to 52% of primary ovarian SCC are positive for HPV.[1,8] Since fewer cases have been reported in literature there is no proper management protocol for these cases. However, the patients are treated with radical surgery followed by a chemotherapy regimen of Paclitaxel and Platinum-based drugs. In 1998, Gamal et al reported a case of primary ovarian SCC that showed a remarkable response to a chemotherapy regimen of Paclitaxel and Cisplatin.[2,6–9,12]

4. Conclusion

Epidermoid cysts of the ovary transforming into SCC is a rare entity which every pathologist should be aware of. Since preoperative diagnosis of ovarian SCC is difficult, histopathological analysis becomes inevitable.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Tobar PR, Assad RA, Nazal ON, Suárez ER. Ovarian squamous cell carcinoma. *J Cancer Res Ther*. 2022;18:777–9.
2. Luo Y, Bian C. Primary squamous cell carcinoma of the ovary. *Front Oncol*. 2022;12:1–6.
3. Abid Z, Fatima M, Zehra D, Abid M, Khan SA. Primary squamous cell carcinoma of the ovary. *Cureus*. 2019;11:1–3.
4. Park JW, Bae JW. Pure primary ovarian squamous cell carcinoma: a case report and review of the literature. *Oncol Lett*. 2015;9(1):321–3.
5. Agarwal S, Pandey P, Ralli M, Singh S. Squamous-Cell Carcinoma Arising from an Epidermoid Cyst of the Ovary and Metastasizing to the Uterus: Report of an Unusual Case with Review of Literature. *J Gynecol Surg*. 2017;33:156–9.
6. Eltabbakh GH, Hempling RE, Recio FO, O'Neill CP. Remarkable response of primary squamous cell carcinoma of the ovary to paclitaxel and cisplatin. *Obstet Gynecol*. 1998;91(5 Pt 2):844–6.
7. Khanfar NM, Arndt CA, Wold LE. Squamous cell carcinoma in the ovary of a 14-year-old girl. *Mayo Clin Proc*. 1996;71(4):380–3.
8. Bak S, Hong JY, Lee JW, Im S, Park DC. Primary squamous cell carcinoma of the ovary accompanied by transition of a mucinous borderline ovarian tumor. *J Int Med Res*. 2023;51(2):1–6.
9. Koufopoulos N, Kokkali S, Antoniadou F, Nasi D, Goudeli C, Terzakis E, et al. EP895 Primary squamous cell carcinoma of the ovary: review of the literature. *Int J Gynecol Cancer*. 2019;29:1776–84.
10. Sharma N, Manimekalai, Sonti S, Meenakshisundarum. A rare case of a giant epidermoid cyst of the ovary. *J Clin Diagn Res*. 2012;6(9):1584–6.
11. Pins MR, Young RH, Daly WJ, Scully RE. Primary squamous cell carcinoma of the ovary: report of 37 cases. *Am J Surg Pathol*. 1996;20(7):823–33.
12. Kikkawa F, Nawa A, Tamakoshi K, Ishikawa H, Kuzuya K, Suganuma N, et al. Diagnosis of squamous cell carcinoma arising from mature cystic teratoma of the ovary. *Cancer*. 1998;82(11):2249–55.

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