CASE REPORT

A case of metachronous colon cancer developing after a primary granulosa cell tumor of ovary

Santosh Kumar Dora, Pariseema Dave, Meeta Mankad, Anusha Kamath

Correspondence: Dr. Santosh Kumar Dora, Gujarat Cancer Research Institute, Department of Gynaecology Oncology, Ahmedabad, Gujarat, India; Emailsantoshdora1@gmail.com

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ABSTRACT

Recurrence after a granulosa cell tumor of ovary is though common, metachronous tumor should always be kept in mind because management of both cancers differ significantly. A 61 years old woman presented with a pelvic mass and enterocutaneous fistula with past history of granulosa cell tumor of the ovary for which surgery and chemotherapy was received 7 years back. Her inhibin B level was 70 pg/ml and CA125 was 7.05 U/ml giving the probable diagnosis of a recurrent granulosa cell tumor of ovary with a complicated enterocutaneous fistula. She underwent exploratory laparotomy with enbloc resection of pelvic mass and the final histopathological examination report came out to be mucinous adenocarcinoma of colon and appendix.

Keywords: Ovary, granulosa cell tumour, metachronous colon, adenocarcinoma.

Granulosa cell tumour (GCT) is a rare variety of ovarian tumour. It comprises of 2-5% of all ovarian malignancy [1]. GCTs have a tendency of late recurrence and on recurrence it has a grave prognosis. Mucinous adenocarcinoma of colon developing after a granulosa cell tumour is the rarest variety of metachronous tumour. We describe it as a rarest combination of mucinous adenocarcinoma of colon developing seven years after primary GCT.

Case report

A 61 years old woman presented with a pelvic mass and enterocutaneous fistula. Her significant past history revealed a granulosa cell tumor of the ovary, for which total abdominal hysterectomy and bilateral salpingoophorectomy followed by staging laparotomy and completion of surgery at a higher centre (Bilateral pelvic lymphadenectomy, para aortic LN sampling, infracolic omentectomy) was done seven years back. Her histopathology slides were reviewed and confirmed to be adult variety of GCT at a tertiary care national institute of India. After that she had also received three cycles of carboplatin and paclitaxel and following which she had a disease free period for about seven years. But subsequently she developed abdominal pain and an abscess over the umbilicus and previous scar. The abscess was drained and the material was sent for histopathological examination but it didn't reveal any malignant cell. Imaging studies with USG

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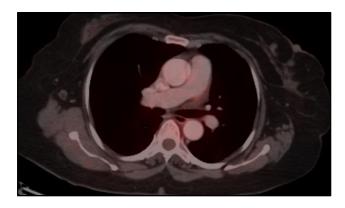


Figure 1: A sinus tract communicating skin surface to the underlying bowel loop

and CECT abdomen and pelvis revealed a hypodense lesion of size $4.5 \, \text{cm} \times 3.3 \, \text{cm} \times 4.4 \, \text{cm}$ in the peritoneal cavity communicating with the umbilicus, caecum and ascending colon. USG guided FNAC from the lesion showed no evidence of malignant cell. But her PETCT (figure-1) showed the same lesion in the infraumbilical area with the possibility of sinus tract

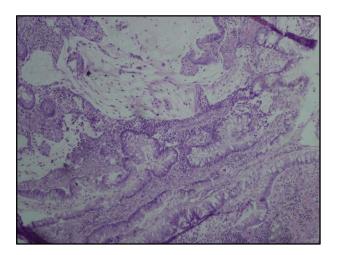


Figure 2: Mucin producing adenocarcinoma of colon

infection (SUV max=10.9). Confusing the picture more her inhibin B level was 70 pg/ml (normal \leq 17.5 pg/ml) and CA125 was 7.05 U/ml giving the probable diagnosis of a recurrent granulosa cell tumor of ovary

with a complicated enterocutaneous fistula. Then she underwent exploratory laparotomy with enbloc resection of pelvic mass, excision of the sinus tract and right hemicolectomy. Right hemicolectomy was done in view of frozen section showed possibility of colonic adenocarcinoma. The final HPE (histopathological examination) report came out to be mucinous adenocarcinoma of colon (figure-2) and appendix. Now she is under follow up and on chemotherapy oxaliplatin and capacitabine.

Discussion

GCT is one of the commonest sex cord stromal cell tumour of the ovary, with a tendency of late recurrence. We have made a provisional diagnosis of recurrent GCT as around 21% of GCT recurs with a median time of recurrence of about 57.6 month as seen in our patient, who had recurrence after a time period of about 7 years [2]. Most of the recurrence is seen in pelvis (70%), 9% in both abdomen & pelvis & 3% in pelvis, abdomen, retroperitoneum [3]. We are probably reporting the first case of metachronous GCT of ovary with adenocarcinoma of colon.

Metachronous tumour are second malignant tumour develop more than 6 months after the primary tumour. Colonic and appendiceal adenocarcinoma developing after granulosa cell tumour is one of the rarest occurrences. Metachronous tumor have been reported in literature from time to time, as adenocarcinoma of colon with carcinoid tumour [4], transitional cell tumor of bladder and adenocarcinoma of colon [5], adenocarcinoma of colon and yolk sac tumor of ovary [6]. The etiologic factors implicated for the development of such metachronous tumour are the primary tumour itself, genetic manipulation, genetic mutation, chemotherapy, radiotherapy etc. There are few hypotheses regarding the development of such type of metachronous tumour, though there is no satisfactory explanation to it [7] - i) Two different cell lines proliferate in a particular time, ii) Two different cell lines proliferate from a single pluripotent stem cell, iii) Two unrelated tumor grow synchronously or metachronously by chance.

But the question which needs further studies is, do these tumours occur by chance or is there a single carcinogenic event which leads to the development of both these tumour...?? Colon cancer is one of the most common malignant tumour of the developed world [8]. Incidence of metachronous colorectal cancer is 0.5-0.9% and the average time taken for its manifestation is 7-11 years. In this case, patient presented with an enterocutaneous fistula with HPE and PET scan report suggestive of infective aetiology makes preoperative diagnosis more difficult. So to diagnose a metachronous tumor we should broaden our differential diagnosis, management of primary metachronous tumor are totally different. GCT has got a good prognosis though the outcome depends mainly on the stage of tumour and capsular rupture. But due to infrequent combination of the two tumours, it is difficult to ascertain the final prognosis of the disease as to which component actually determines the final outcome. As in this case beside the rarest incidence of tumour, the presentation of the tumour was also peculiar.

Conclusion

GCT is known for its late recurrence and they should be followed up with appropriate tumour markers and imaging. But clinical awareness and recognition of a secondary malignancy should always be kept in mind. Our case highlights the importance of such possibility and spread awareness among gynaecology-oncologist and medical oncologist. In conclusion recognition of both components of tumour is important as it dictates the overall prognosis of the patient.

Conflict of interest: None. Disclaimer: Nil.

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Santosh Kumar Dora¹, Pariseema Dave², Meeta Mankad³, Anusha Kamath⁴

^{1,2,3,4} Gujarat Cancer Research Institute(GCRI), Department of Gynaecology Oncology, Ahmedabad, Gujarat, India.