Volume 19 Issue 2
Originally issued as K1

Global Journal of Medical Research: K
Interdisciplinary
Volume 19 Issue 1 Version 1.0 Year 2019
Type: Double Blind Peer Reviewed International Research Journal
Publisher: Global Journals
Online ISSN: 2249-4618 & Print ISSN: 0975-5888

Rare Primary Paraovarian Adenocarcinoma in Postmenopausal Woman: Case Report and Review

By Dr. Anjani Dixit & Dr. Shanti Jayaseelan

Abstract- Malignant paratubal cancers have extremely rare occurrence and have not been sufficiently described and discussed in literature. We describe case of an 80 years old gravida 5 para 4 postmenopausal woman presented with pain in lower abdomen and spotting per vagina. USG followed by MRI showed large mid pelvic right adnexal complex predominantly cystic lesion of 8.7x7.1x5.9 cm size with small exophytic solid component (3.1x2.8x2.6 cm). Postoperatively it was diagnosed as a case of high grade paratubal adenocarcinoma (stage T1N0M0). The patient received 3 cycles of adjuvant chemotherapy with carboplatin and docetaxel. Patient has had no recurrence till date (20 months post treatment).

Keywords: *paratubal adenocarcinoma, adnexal complex, primary tumour, broad ligament.*

GJMR-K Classification: *NLMC Code: WP 460*

RAREPRIMARYPARAOVARIANADENOCARCINOMAINPOSTMENOPAUSAL WOMANCASEREPORTANDREVIEW

Strictly as per the compliance and regulations of:

© 2019. Dr. Anjani Dixit & Dr. Shanti Jayaseelan. This is a research/review paper, distributed under the terms of the Creative Commons Attribution-Noncommercial 3.0 Unported License http://creativecommons.org/licenses/by-nc/3.0/), permitting all non commercial use, distribution, and reproduction inany medium, provided the original work is properly cited.

Rare Primary Paraovarian Adenocarcinoma in Postmenopausal Woman: Case Report and Review

Dr. Anjani Dixit [α] & Dr. Shanti Jayaseelan [σ]

Abstract- **Malignant paratubal cancers have extremely rare occurrence and have not been sufficiently described and discussed in literature. We describe case of an 80 years old gravida 5 para 4 postmenopausal woman presented with pain in lower abdomen and spotting per vagina. USG followed by MRI showed large mid pelvic right adnexal complex predominantly cystic lesion of 8.7x7.1x5.9 cm size with small exophytic solid component (3.1x2.8x2.6 cm). Postoperatively it was diagnosed as a case of high grade paratubal adenocarcinoma (stage T1N0M0). The patient received 3 cycles of adjuvant chemotherapy with carboplatin and docetaxel. Patient has had no recurrence till date (20 months post treatment).**

Keywords: paratubal adenocarcinoma, adnexal complex, primary tumour, broad ligament.

Key Messages: Primary paratubal adenocarcinoma is an extremely rare case. To the best of our knowledge, we here report the oldest patient (80 years) with this condition. She presented with vaginal spotting which is a close mimic of endometrial cancer in this age. Diagnosis and management together with review of literature are discussed. It is observed that due to rarity of the case, there is no consensus on management. Further studies and reporting are recommended.

I. Introduction

Primary paraovarian malignancy is an extremely rare condition, so much so that very little is known about their epidemiology, biological behaviour, prognosis, protocols for diagnosis and management. The incidence of paraovarian malignancy is also not known[16]. Very few cases have been reported in literature till date. We hereby present, to the best of our knowledge, the oldest patient in literature with paraovarian malignancy. Diagnosis and management together with review of literature are discussed.

II. Case Report

An eighty years old gravida 5 para 4 postmenopausal woman presented with continuous pain in lower abdomen for last one and a half year and vaginal spotting for last 4 days. Patient was menopausal for last 30 years. On pelvic examination, cervix and vagina were assessed to be normal, uterus was normal and anteverted, a cystic mass of approximately 6x6 cm in size was felt close to uterus, and POD was free. USG was suggestive of thin walled anechoic cystic mass on right side, 8x6x6 cm with few internal septae. MRI pelvis showed large midpelvic right adnexal complex predominantly cystic lesion of size 8.7 x 7.1 x5.9 cm with small exophytic solid component (3.1x2.8x2.6 cm). The exophytic solid component was focally indenting anterior rectal wall with no apparent infiltration. Endometrial thickness was 1.7 mm, smooth regular margins and fluid – fluid levels were seen. Endometrial curettage was done and histopathology report showed senile endometrium. Her serum CA125 level was normal.

Patient underwent exploratory laparotomy. No ascites or peritoneal implants were seen. Peritoneal wash was sent for cytological examination. Both the ovaries, fallopian tubes and the uterus were normal in appearance. A large paraovarian cystic mass measuring 8x6x4cm was seen on right side with right fallopian tube stretched over it. TAH with BSO with cystectomy was done. Full biochemical and radiological examinations showed no evidence of metastasis or any other disease process. We staged the primary paraovarian tumour as T1aN0M0. Five weeks later, patient underwent 3 cycles of adjuvant chemotherapy with carboplatin and docetaxel. Patient is doing well in follow up visits till date (20 months post treatment).

III. Pathology

On postsurgical examination, peritoneal fluid cytology showed no evidence of malignancy. On macroscopic examination, paraovarian cyst was found to be uniloculated, filled with thin hemorrhagic fluid and a firm, greyish white nodule measuring 3x2.5x2 cm was seen in the wall of the cyst, which on cut surface was grey white, granular with large areas of necrosis. Rest of the cyst wall had smooth inner surface with foci of hemorrhages. No tumour was found in the bilateral ovaries and fallopian tubes.

Author α: MBBS, DNB, Senior Resident, Department of Obstetrics & Gynaecology, Holy Family Hospital, 143A, First floor, ShahpurJat, New Delhi-110049. e-mail: doc.anjani@gmail.com
Author σ: MBBS, MD, Senior Consultant, Department of Obstetrics & Gynaecology, Holy Family Hospital, Okhla road, New Delhi- 110049. e-mail: shanthijayaseelan@gmail.com

© 2019 Global Journals

Microscopic examination of the right adnexal mass revealed malignant tumour with solid and tubulocystic pattern with cells lining the tubules having hobnail appearance. There was marked pleomorphism in the neoplastic cells in the solid area suggestive of high grade serous adenocarcinoma from paraovarian cyst (Fig. 1 and 2). Large areas of necrosis and stromal lymphocytic infiltration were seen. Sections of the cyst wall showed columnar to cuboidal to flattened cell lining with underlying fibrosis and infiltration by hemosiderin laden macrophages. There was no lymphovascular space invasion. Endomyometrium showed senile cystic atrophy with atrophic myometrium. Bilateral fallopian tubes and ovaries were unremarkable.

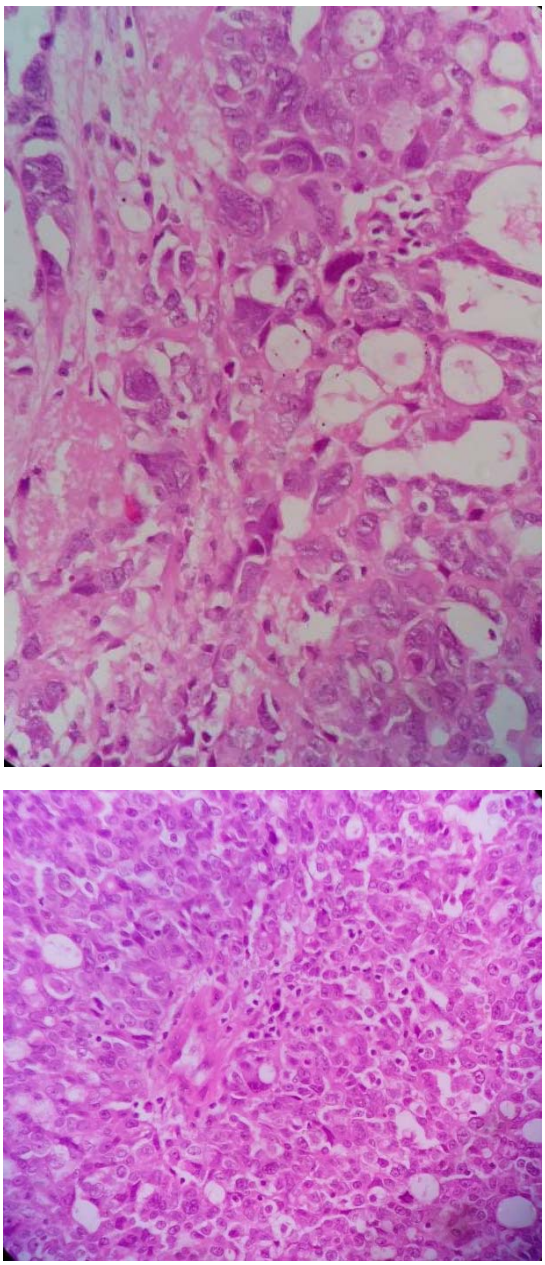


Fig. 1 and Fig. 2: High grade serous adenocarcinoma showing papillary structure with marked pleomorphism and frequent mitosis

IV. Discussion

Secondary tumours in broad ligament are not uncommon but primary tumours are rare. Commonly seen primary tumours are leiomyomata, serous and papillary cystadenoma of borderline malignancy but primary malignant tumours are very rare[9]. Primary broad ligament carcinoma as mesonephroma was first reported by *Schiller* in 1939[1].

In our case primary tumour was located in or on the broad ligament and completely separated from ipsilateral ovary, fallopian tube, and uterus. This satisfies the criteria given by *Gardner et al* in 1957 to diagnose primary carcinoma of broad ligament origin[4].

The age range in reported cases of broad ligament malignant tumours is 13 to 80 years (median 43 years) and the age range of borderline tumours is 28-38 years (mean 32.6 years) with 43.4% below the age of 40 years. To the best of our knowledge, ours (80 years) is the oldest case reported in the literature. In contrast, mean age of ovarian cancer is 63 years, being rare in women below 40 years[18] and mean age of borderline ovarian tumours is 10 years lower[19].

Five were clear cell carcinoma (20.8%), two were papillary adenocarcinoma (8.3%), four were endometroid (16.6%), five including ours were serous adenocarcinoma (20.8%), one was mucinous carcinoma (4.1%), and one was well differentiated adenocarcinoma (4.1%) and five were borderline serous cystadenoma (20.8%) *(Table 1)*.

Most common presenting symptoms reported are lower abdominal pain and pelvic discomfort[15] like in our case. Moreover, our patient also presented with vaginal spotting. Although most common cause for vaginal spotting in post-menopausal age group is senile endometritis[20], there is a possibility of associated endometrial carcinoma as mentioned by *Aslani et al*[9]. Hence it is important to rule out endometrial cancer in scenarios of vaginal spotting for postmenopausal women. In our case endometrial cancer was ruled out by endometrial curettage. It is to be noted that presentation of broad ligament tumour can be an incidental finding as well[9]. There is a 1.4 % to 3.8% chance of synchronous tumour of ovary and endometrium[20] but similar data about broad ligament tumour is not known due to rarity of cases. All the reported cases of broad ligament tumour *(Table 1)* including borderline variety were unilateral, in contrast to ovarian tumour which are 25% bilateral[21].

For fourteen cases (58%) including ours, patients were diagnosed in stage 1 of broad ligament tumour. Two cases (8.3%) were diagnosed in stage 2, while three cases (12.5%) were diagnosed in stage 3. Staging was not known for the remaining five cases (20.8%). It is to be noted that majority of the cases, including our case, were diagnosed in stage 1. Possible

© 2019 Global Journals

reason for the same is that tumours are encapsulated between the sheets of broad ligament. Thus rupture as well as aggressive progression is prevented due to lack of vasculature[22].

Management of broad ligament tumour is often done in a similar way as that of ovarian cancer because of similarities in histology and histogenesis, i.e. commonality in coelomic epithelium[10]. Post-surgical adjuvant treatment is not established as the standard procedure due to lack of evidence. It is recommended to use same principles as followed in the management of ovarian cancer[15]. Of the 24 cases, surgery was the singular mode of treatment in eleven cases (46%), surgery followed by chemotherapy in eight cases including our case (33%), surgery followed by radiotherapy in four cases (17%) while only radiotherapy was used in one case (4%).

Table 1: Summarised review of literature

S. No.	Author (year)	Age (years)	Size (cm)	Pathological diagnosis	Treatment	Follow up	stage
1	*Schiller*[1] (1939)	43	Child's head	Clear cell	TAH+BSO, Radiotherapy	Live, 24 months	1
2	*Lennox*[2] (1952)	45	7x6.5x6	Papillary adenocarcinoma	TAH+BSO, Radiotherapy	Live, 10 months	1
3	*Telium*[3] (1954)	62	Fist size	Clear cell	Enucleation	Not known	2
4	*Telium*[3] (1954)	32	10x8x8	Clear cell	Enucleation	Not known	1
5	*Gardner*[4] (1957)	50	-	Well differentiated adenocarcinoma	Radiotherapy	Live,27 months	--
6	*Merril*[5] (1959)	70	13	Papillary adenocarcinoma	TAH+BSO, Radiotherapy	Live,12 months	2
7	*Czernobilsky*[6] (1972)	29	5x4x4	LMP Serous	TAH+BSO	Not known	1
8	*Genadry*[7] (1977)	13	9x6	Serous adenocarcinoma	Adnexectomy	Live, 60 months	--
9	*Genadry*[7] (1977)	38	9x6	LMP Serous	Adnexectomy	Live, 60 months	--
10	*Genadry*[7] (1977)	36	9x6	LMP Serous	Adnexectomy	Live, 60 months	--
11	*Genadry*[7] (1977)	28	9x6	LMP Serous	Adnexectomy	Not known	--
12	*Clark*[8] (1979)	29	8x7x2	Mucinous adenocarcinoma	TAH+BSO+ omentectomy	Not known	1
13	*Aslani*[9] (1989)	51	10x6x6	Clear cell	Excision+BSO+ omentectomy, Radiotherapy	Live, 7 months	1
14	*Aslani*[9] (1989)	29	6x6x5	Endometroid	Excision+TAH+BSO, inguinal and paraaortic lymph node sampling, Chemotherapy	Live, 18 months	1
15	*Aslani*[9] (1989)	69	11x8	Endometroid	Excision of tumor + TAH+BSO, Chemotherapy	Live, 12 months	1
16	*Aslani*[9] (1989)	34	4.5x4x3.5	Endometroid	TAH+BSO+ plevic and paraaortic lymph node dissection+ omentectomy, Chemotherapy	Live, 6 months	1
17	*Altaras*[11] (1990)	76	12x9x8	Serous	TAH+BSO	Live, 53 months	1

© 2019 Global Journals

18	*Itani*[11] (2001)	54	4.7x5.7	Serous	TAH+BSO+ pelvic and periaortic lymphadenectomy+ omentectomy, chemotherapy	Live, 18 months	1
19	*Vaysse*[12] (2009)	44	16x14x5	Endometroid	TAH+BSO+ Pelvic and paraaorticlymphadnectomy+ omentectomy	Live, 36 months	1
20	*Kaur*[13] (2011)	37	13x8x5	Endometroid	TAH+BSO+ omentectomy+ plevic lymphadenectomy, chemotherapy	Live, 3 months	3
21	*Jong-Hyun Kim*[14] (2013)	32	17×12×10	Serous LMP	RSO+omentectomy +appendectomy+ peritoneal biopsies +right pelvic lymphadenectomy	Live, 24 months	1
22	*Miyoshi*[15] (2015)	71	7.4x6.4x5.2	Serous	Modified radical hysterectomy+BSO + omentectomy, chemotherapy	Live, 5 months	3
23	*Miyoshi* [15] (2015)	43	4x3.7x3.9	Clear cell	Modified radical hysterectomy+BSO + omentectomy + pelvic lymphadenectomy, chemotherapy	Live, 3 months	3
24	Our Case (2018)	80	8x6x4	serous	TAH+BSO, Chemotherapy	Live, 24 months	1

Abbreviations: TAH- Total abdominal hysterectomy, BSO- Bilateral salpingoophorectomy, RSO- Right salpingoophorectomy,

V. Conclusion

Survival rates and prognostic factors are inconclusive due to rarity of the tumour, lack of uniformity in treatment modalities and improper reporting. Similar to cases of ovarian cancer, follow up is important to look for relapse or any residual disease left after adjuvant therapy.

We recommend that such rare cases be reported in literature so that consensus on diagnosis and management can be evolved and established.

References Références Referencias

1. Schiller W. Mesonephromaovarii. Am J Cancer 1939; 35:1-21.
2. Lennox B, Meagher D J. Parovarian carcinoma. BJOG: An International Journal of Obstetrics & Gynaecology. 1952 Dec 1; 59(6):783-5.
3. Telium G. Histogenesis and classification of mesonephric tumours of the female and male genital system and relationship to benign so called adenomatoid tumours (mesothelioma). Acta Pathol Microbiol Scand. 1954; 34: 431.
4. Gardner GH, Greene RR, Peckham B. Tumors of the broad ligament. Obstetrical & Gynecological Survey. 1957 Oct 1; 12(5):775-7.
5. Merrill J A. Carcinoma of the broad ligament. Obstetrics & Gynecology. 1959 Apr 1; 13(4): 472-6.
6. Czernobilsky B, Lancet M. Broad ligament adenocarcinoma of Müllerian origin. Obstetrics & Gynecology. 1972 Aug 1; 40(2):238-42.
7. Genadry R, Parmley T, Woodruff J D. The origin and clinical behaviour of the paraoavrian tumour. Am J ObstetGynecol 1977; 129: 873-80.
8. Clark J E, Wood H, Jaffurs W J, Fabro S. Endometrioid-type cystadenocarcinoma arising in the mesosalpinx. Obstetrics and gynecology. 1979 Nov; 54(5):656-8.
9. Aslani M, Scully R E. Primary carcinoma of the broad ligament. Report of four cases and review of the literature. Cancer. 1989 Oct 1; 64(7):1540-5.
10. Altaras M M, Jaffe R, Corduba M, Holtzinger M, Bahary C. Primary paraovariancystadenocarcinoma: clinical and management aspects and literature review. Gynecologic oncology. 1990 Aug 1; 38(2): 268-72.

© 2019 Global Journals

11. Itani Y, Itoh K, Adachi S, Koyama M, Kimura T, Chin R, Takemura T. Malignant epithelial tumor of unknown origin of the broad ligament. Archives of gynecology and obstetrics. 2002 Dec 1; 267(2): 113-6.
12. Vaysse C, Capdet J, Mery E, Querleu D. Paratubalendometrioidcystadenocarcinoma: case report and review. Eur J GynaecolOncol. 2009 Jan 1; 30(4):443-5.
13. Kaur N, Kaushik R, Gulati A, Kaushal V, Bindra R. Primary endometrioid carcinoma of the broad ligament: a rare case report. The Journal of Obstetrics and Gynecology of India. 2014 Feb 1; 64(1):70-2.
14. Kim JH, Cho DH. Primary borderline parovarian tumor in pregnancy. Obstetrics &gynecology science. 2015 Nov 1; 58(6):533-6.
15. Miyoshi A, Miyatake T, Hara T, Komiya S, Komura N, Tanaka A, Kanao S, Takeda M, Mimura M, Nagamatsu M, Yamasaki M. Rare Primary Adenocarcinoma of the Broad Ligament: Report of Two Cases and a Literature Review. International journal of surgical pathology. 2016 Aug; 24(5): 436-42.
16. Yoonessi M.: "Carcinoma of the fallopian tube". Obstet. Gynecol. Surv., 1979, 34, 257.
17. Savelli L, Ghi T, De Iaco P, Ceccaroni M, Venturoli S, Cacciatore B. Paraovarian/paratubal cysts: comparison of transvaginal sonographic and pathological findings to establish diagnostic criteria. Ultrasound in obstetrics & gynecology. 2006 Sep 1; 28(3):330-4.
18. American Cancer Society.(no date) Ovarian Cancer Risk Factors [online] [accessed on 22 july 2018] https://www.cancer.org/cancer/ovarian-cancer/causes-risks-prevention/risk-factors.html
19. Green A E, Huh W K. Borderline Ovarian Cancer: Borderline Tumor Overview. Online document at: www.emedicine.medscape.com/Accessed August. 2012 Aug 18; 18.
20. Berek J S. Berek & Novak's Gynecology. 15th ed. Philadelphia, USA: Lippincott Williams & Wilkins; 2012.
21. Micci F, Haugom L, Ahlquist T, Abeler V M, Trope C G, Lothe R A, Heim S. Tumor spreading to the contralateral vary in bilateral ovarian carcinoma is a late event in clonal evolution. Journal of oncology. 2010; 2010.
22. Handa Y, Kato H, Kaneuchi M, Saitoh Y, Yamashita K. High-grade broad ligament cancer of müllerian origin: immunohistochemical analysis of a case and review of the literature. International Journal of Gynecological Cancer. 2007 May; 17(3):705-9.

© 2019 Global Journals