International Journal of Clinical and Diagnostic Pathology

ISSN (P): 2617-7226
ISSN (E): 2617-7234
www.patholjournal.com
2019; 2(1): 321-324
Received: 11-05-2019
Accepted: 18-05-2019

Chiragkumar B Menapara
Assistant Professors, Department of Pathology, GMERS Medical College & Hospital, Near Majewadi Gate, Paddock Road, Junagadh, Gujarat, India

Mayur J Kokani
Assistant Professors, Department of Pathology, GMERS Medical College & Hospital, Near Majewadi Gate, Paddock Road, Junagadh, Gujarat, India

Correspondence
Mayur J Kokani
Assistant Professors, Department of Pathology, GMERS Medical College & Hospital, Near Majewadi Gate, Paddock Road, Junagadh, Gujarat, India

Squamous cell carcinoma arising in an ovarian mature cystic teratoma: A case report

Chiragkumar B Menapara and Mayur J Kokani

DOI: https://doi.org/10.33545/pathol.2019.v2.i1e.48

Abstract
Pure primary squamous cell carcinoma (SCC) of the ovary is extremely rare. SCC can also arise in a mature cystic teratoma or dermoid cyst, in an ovarian endometrioma or in a brenner's tumor, but such malignant transformations are also quite rare. Development of SCC in a mature cystic teratoma (MCT) has been reported sporadically in less than 2% cases only.
Here in our present study, we are reporting such a rare case of 65 years old postmenopausal woman who was diagnosed as having squamous cell carcinoma developed in a mature cystic teratoma of the ovary at our institute. This diagnosis was given on the basis of clinical (patient's history and examination), radiological (CT scan) and pathological (histopathology report) findings. We are also going to discuss other similar cases with review of literature. The only aim of this present study is to share our experience of such rare diagnosis with others.

Keywords: Mature cystic teratoma, squamous cell carcinoma, malignant transformation

Introduction
Development of squamous cell carcinoma (SCC) as a pure (de novo) or primary malignant tumor in the ovary is an aggressively rare entity [1]. The pure variety arises from metaplasia of surface epithelium of ovary and it is the rarest type. Development of SCC in a mature cystic teratoma (MCT) or dermoid cyst, in an ovarian endometrioma or in a brenner's tumor is also quite rare. [2] Malignant transformation develops in less than 2% of MCT and approximately 75% of them are squamous cell carcinomas (SCC), followed by adenocarcinoma and sarcoma or melanoma. [3, 4] The mechanism of such malignant transformation has not been completely understood yet and it is difficult to diagnose the ovarian SCC arising in a teratoma preoperatively, due to its rarity, vague symptoms, and clinically aggressive course. [5]
In this present study, we are reporting such a rare case of 65 years old postmenopausal woman who was presented and admitted at the gynecology department of our institute with complaints of abdominal enlargement and pain. On radiological examination by CT scan, left sided ovarian cystic teratoma was diagnosed and on the basis of both clinical and radiological findings, surgical removal of tumor was performed and the specimen was further sent to the histopathology laboratory for final diagnosis.
At laboratory, we performed both gross and microscopic examination of the specimen itself and we took sections selectively from thickened parts of the cyst wall. Finally, we came to the diagnosis of squamous cell carcinoma developed in a mature cystic teratoma. Many similar rare cases have been reported till now and the only aim of this present study is to share our experience of such rare diagnosis with others.

Case report
A 65 years old post-menopausal woman presented with painful enlargement of abdomen that was not associated with any weight-loss, loss of appetite, bowel-bladder disturbances, etc. On examination, a palpable lump was identified in abdomen arising from pelvic region, around 24 week size and not associated with free fluid inside the peritoneal cavity. CT scan revealed a large well-defined fat density space occupying lesion (SOL), located in pelvis, predominantly in midline and slightly in left para-median position, arising from ovary, approximately measuring 8 x 8 x 7 cm^3 in size and having partially solid and partially cystic appearance with variable amount of opacities.

Few septations as well as foci of calcification were also noted within the lesion. Patient's serum CA125 level was 12.58 U/ml (within normal range or limit). So, provisional and pre-operative diagnosis of Mature Cystic Teratoma (MCT) or Dermoid Cyst was established.

Gross examination of surgical pathology specimen revealed a grayish white cystic soft tissue mass approximately measuring 8 x 8 x 7 cm^3 in size with smooth and uniform outer surface. On cutting, putty like foul smelling material admixed with a tuft of hair was identified. Cyst-wall thickness was variable and thickened areas were quite identifiable. (Figure no.1)

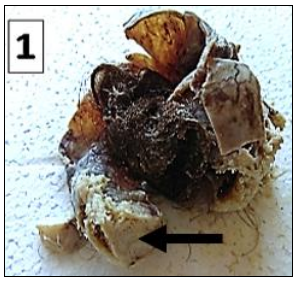

Fig 1: Gross appearance of specimen with a tuft of hair and thickened part of the cyst wall (black arrow)

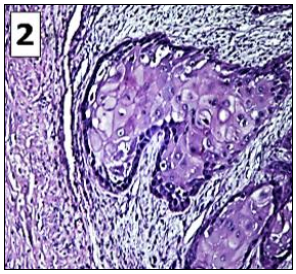

Fig 2: Photomicrograph showing normal ovarian stromal tissue admixed with foci or islands of malignant looking squamous epithelial cells (H & E stained section, low power magnification)

Microscopic examination of H & E stained sections revealed normal ovarian stromal tissue admixed with islands of malignant looking squamous epithelial cells (Figure no. 2 & 3). At places, excessive keratinization and keratin pearl formation were also evident (Figure no.4). Occasionally, normal looking stratified squamous epithelium and dermal appendages like sebaceous glands, hair follicles, etc. were also evident. Finally, the diagnosis of Squamous Cell Carcinoma (SCC) with Mature Cystic Teratoma (MCT) was established.

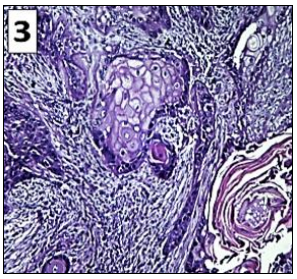

Fig 3: Photomicrograph showing normal ovarian stromal tissue admixed with foci or islands of malignant looking squamous epithelial cells (H & E stained section, low power magnification)

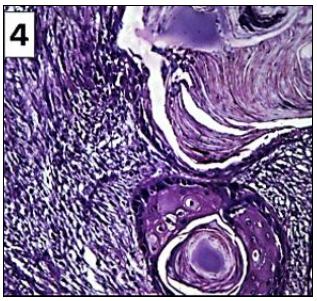

Fig 4: Photomicrograph showing foci or islands of malignant looking squamous epithelial cells with excessive keratinization and keratin pearl formation (H & E stained section, low power magnification)

Discussion

At our department, we have reported a total number of 60 cases of ovarian neoplasms over a period of 4 years. Out of them, mature cystic teratoma or dermoid cyst constitutes a total number of 15 cases (25%). Age-wise distribution of these cases is mentioned below in table no.1.

Table 1: Age-wise distribution of cases of Mature Cystic Teratoma

Age-group (in years)	Number of cases
20-29	02
30-39	05
40-49	05
50-59	01
60-69	02
TOTAL	15

So, we can say that mature cystic teratoma can occur at any age. It can also be seen among post-menopausal & elderly females (above 60 years). In our present case-study, age of the patient herself is 65 years. Comparison of age of occurrence with that of other similar studies is mentioned below in table no. 2.

Table 2: Comparison of age of occurrence of malignant transformation in mature cystic teratoma (MCT) among different studies

Name of the study	Year of publication	Numbers of cases reported	Age of occurrence
Santwani PM *et al.* [6]	2008	01	40 years
Gupta V *et al.* [10]	2009	02	30 years, 65 years
Bashyal R *et al.* [3]	2012	03	33 years, 51 years, 60 years
Suna Avc1 *et al.* [9]	2012	01	52 years
Gupta N *et al.* [4]	2014	01	65 years
Ranu Patni [11]	2014	01	53 years
Mardi K *et al.* [12]	2014	01	68 years
Yarmohammadi H *et al.* [13]	2014	01	48 years
Rekhi B *et al.* [14]	2015	12	Median age = 49 years
Sharma S *et al.* [1]	2015	01	66 years
Abhilasha N *et al.* [8]	2016	10	Median age = 53.5 years
Srivastava H *et al.* [2]	2017	01	30 years
Goudeli C *et al.* [7]	2017	01	54 years

Indulkar ST *et al.* [5]	2018	01	40 years
Maharjan S [15]	2019	01	43 years
Present Study	2019	01	65 years

From the data mentioned above in table no.2, it is obvious that malignant transformations in a mature cystic teratoma are commonly found among middle aged & elderly females (around & beyond 50 years of age). In other words, we can say that peri menopausal & post-menopausal women are more prone to develop such transformations.

In our present study, painful enlargement or distension of abdomen is the presenting complaint of the patient herself. Similar complaints have been reported in other case studies also like those done by Bashyal R *et al* [3], by Gupta N *et al* [4], by Santwani PM *et al* [6], etc. Some authors also reported other complaints like constipation, loss of appetite, loss of weight, low grade fever, etc. (e.g. Goudeli C *et al* [7]) Yarmohammadi H *et al* [13] reported a similar case presented with complaints of nausea, non-bloody non-bilious vomiting, diarrhoea, abdominal cramps & night sweats with final diagnosis of small bowel obstruction done by tumor itself. Indulkar ST *et al* [5] reported a similar case of 40 years old female who presented with complaints of acute abdominal pain and dysmenorrhea. From all these findings, it is obvious that cases of squamous cell carcinoma developed in a mature cystic teratoma clinically represent with vague or non-specific features. Abdominal enlargement, mass in abdomen, discomfort or pain, menstrual irregularities, pressure symptoms like constipation & bowel obstruction, etc. can be seen in these cases.

Tumor size itself revealed by radiological investigations like CT scan, MRI, etc. and by gross examination of surgical pathology specimens also gives some information in such cases that become helpful in their final diagnosis. Comparative evaluation of tumor size of different studies is mentioned below in table no.3.

Table 3: Comparative Evaluation of Tumor Size of Different Studies

Name of the study	Numbers of cases reported	Size of the tumor itself
Santwani PM *et al* [6]	01	12 x 8 cm^2
Gupta V *et al* [10]	02	12 cm & 16 cm in their maximum dimensions
Bashyal R *et al* [3]	03	15 x 12 x 11 cm^3, 14 x 9 x 5 cm^3 & 10 x 9 x 4 cm^3
Suna Avc1 *et al* [9]	01	12 x 7.5 cm^2
Gupta N *et al* [4]	01	15 x 13 x 10 cm^3
Mardi K *et al* [12]	01	10 x 6 x 4 cm^3
Yarmohammadi H *et al* [13]	01	10.3 x 7.6 cm^2
Goudeli C *et al* [7]	01	22 x 18 x 12 cm^3
Indulkar ST *et al* [5]	01	10 x 8 x 5 cm^3
Maharjan S [15]	01	10 x 7 xm^2
Present Study	01	8 x 8 x 7 cm^3

From data mentioned above in table no.3, we can say that tumor size gives important information and it can lead to the diagnosis of malignant transformation in certain cases. In all cases except our present one, the size of the tumor itself measures 10 cm or more in maximum dimension. Kikawa *et al* [16] in his study observed a total number of 37 cases of squamous cell carcinoma referred to their center in 17-year period. A total of 92 patients with mature cystic teratoma (benign) were observed over their study of 3 year-period. The mean size of malignant dermoid was 152.3 mm (15.23 cm) compared to 88.4 mm (8.84 cm) in benign dermoids.

Apart from the size of the tumor, variable cyst wall thickness also gives some clue for the diagnosis of malignant transformation in ovarian teratoma. In our present case also, some thickened or solid parts in the cyst wall were identified on gross examination of surgical pathology specimen. Other studies like those done by Bashyal R *et al* [3], by Gupta N *et al* [4], by Gupta V *et al* [10], by Santwani PM *et al* [6] and by Maharjan S [15] also show similar findings in gross examination of the tumor specimen itself.

Finally, we can say that the teratomas with larger size & variable cyst-wall thickness with thickened or solid area (parts) grossly visible in the cyst-wall have maximum chance of malignant transformations. Sections selectively taken from such thick or solid areas may reveal malignancies (most commonly SCC) on microscopic examination.

Serum level of coelomic antigen (CA) 125 may or may not be elevated in the cases of malignant transformation in mature cystic teratoma. In our present study, it is 12.58 U/ml. In other similar studies done by Bashyal R *et al* [3], by Goudeli C *et al* [7], by Suna Avc1 *et al* [9] and by Maharjan S [15], they are 24.19 U/ml, 37.50 U/ml, 57.00 U/ml and 10.70 U/ml respectively. Gupta V *et al* [10] also reported two cases of malignant transformations in ovarian teratoma with serum CA 125 levels of 40 U/ml & 50 U/ml respectively. Ranu Patni [11] reported a markedly elevated level of CA 125 (143 U/ml). Normal value of serum CA 125 is less than 35 U/ml [3, 7]. So we can say that serum CA 125 level may or may not become helpful in the diagnosis of each and every case of malignant transformation in ovarian teratoma. Other tumor markers useful for the same purpose include CEA and CA 19-9.

Microscopic findings are almost similar in all the cases of squamous cell carcinoma (SCC) developed in a mature cystic teratoma (MCT) and include islands of malignant looking squamous epithelial cells, excessive keratinization & keratin pearl formation (in cases of well differentiated neoplasms only). These features can be easily seen if sections are selectively taken from thickened or solid parts of the cyst wall itself.

Conclusion

Development of squamous cell carcinoma (SCC) in ovaries as a pure primary tumor is extremely rare. Ovarian tumors like mature cystic teratoma (MCT) may show malignant transformations in certain cases and the commonest among them is squamous cell carcinoma. Such transformations are also rare and have been reported sporadically (<2%) till now. Clinical, Radiological & other similar findings like serum CA 125 level may help in the diagnosis of some of these cases but final diagnosis always depends upon gross & microscopic evaluation of surgical pathology specimens. In other words, we can say that the diagnosis of SCC in a teratoma is more or less post-operative in nature, because pre-operative diagnosis may or may not be possible in all such cases.

References

1. Sonam Sharma, Charanjeet Ahluwalia, Sufian Zaheer, Ashish Kumar Mandal. An aggressive rare entity-Pure de novo primary squamous cell carcinoma of ovary: Review of literature and a case report. Clin Cancer Investig J. 2015; 4:682-6.

2. Himsweta Srivastava, Sneha Shree, Kiran Guleria, Usha R Singh. Pure Primary Squamous Cell Carcinoma of Ovary-A Rare Case Report. J Clin Diagn Res. 2017; 11(5). QD01-QD02.
3. Bashyal R, Lee MC. Squamous Cell Carcinoma arising in Ovarian Mature Cystic Teratoma: report of three cases. Journal of Pathology of Nepal. 2012; 2:248-250.
4. Gupta N, Gupta T, Jindal S. Squamous Cell Carcinoma arising in Mature Cystic Teratoma of the Ovary. NJOG. 2014; 17(1):61-63.
5. Shreeya Taresh Indulkar, Manisha Khare S, Vinaya Shah B, Archana Khade L. Squamous Cell Carcinoma arising in Mature Teratoma of the Ovary masquerading as Abdominal Tuberculosis. J Mid-life Health. 2018; 9:44-6.
6. Santwani PM, Trivedi DP, Vachhani JH, Trivedi NJ. Co-existence of squamous cell carcinoma with dermoid cyst of ovary. Indian J Pathol Microbiol. 2008; 51:81-2.
7. Goudeli C, Varytimiadi A, Koufopoulos N, Syrios J, Terzakis E. An ovarian mature cystic teratoma evolving in squamous cell carcinoma: A case report and review of the literature. Gynecol Oncol Rep. 2017; 19:27-30.
8. Abhilasha N, Bafna UD, Pallavi VR, Rathod PS, Krishnappa S. A review of squamous cell carcinoma arising in mature cystic teratoma of the ovary. Indian J Cancer. 2016; 53:612-4.
9. Avc1 S, Selcukbiricik F, Bilici A, Ozkan G, Ozagar1 AA, Borlu F. Case report-squamous cell carcinoma arising in a mature cystic teratoma. Case Rep. Obstet. Gynecol, 2012.
10. Vishwanath Gupta, Neelam Sood. Squamous Cell Carcinoma arising in a mature cystic teratoma. Ind. J Pathol Microbiol. 2009; 52(2):271-3.
11. Ranu Patni. Squamous cell carcinoma arising in mature cystic teratoma of ovary. J Mid-life Health. 2014; 5(4):195-7.
12. Kavita Mardi, Sudershan Sharma. Squamous cell carcinoma arising in an ovarian mature cystic teratoma. Clin Cancer Investig J. 2014; 3:96-8.
13. Yarmohammadi H, Mansoori B, Wong V, Tacher V, Wilkins LR, Pavlidakey PG *et al*. Squamous cell carcinoma arising from ovarian mature cystic teratoma and causing small bowel obstruction. J Can Res Ther. 2014; 10:770-2.
14. Rekhi B, Parikh P, Deodhar KK, Menon S, Maheshwari A, Kerkar R *et al*. Squamous carcinoma coexistent with teratoma of ovary: A clinicopathological study of 12 cases diagnosed over a 10-year period at a tertiary cancer referral center. J Can Res Ther. 2015; 11:211-5.
15. Sushna Maharjan. Mature cystic teratoma of ovary with squamous cell carcinoma arising from it. Clin Case Rep. 2019; 7:668-71.
16. Kikawa F, Nawa A, Tamakashik, *et al*. Diagnosis of squamous cell carcinoma arising from mature cystic teratoma of ovary Cancer. 1998; 82:2249-55.