MODESTUM
OPEN ACCESS

Case Report

Eur J Gen Med 2018;15(1):33-36
ISSN:1304-3889 eISSN:1304-3897
https://doi.org/10.29333/ejgm/81732

Xanthogranulomatous inflammation of the ovary with the uterine adenomatoid tumor in a patient with diabetes mellitus: A case report

Özlem Şengül[1], Gülcan Dauletkazin[2], Levent Keskin[2], Sibel Orhun[3], Filiz Avşar[1]

ABSTRACT

Adenomatoid tumor of the uterus and xanthogranulomatous inflammation of the ovary are very rare lesions. This case report is to document both lesions in a patient with diabetes mellitus. A postmenopausal patient, ultrasonographic examination revealed 72*42mm cystic lesion in adnexial region and 120*40mm sized collection of intraabdominal fluid .The patient had diabetes mellitus for 6 years. Total abdominal hysterectomy and bilateral salphingooferectomy were performed. The pathology report came out as uterin adenomatoid tumor, ovarian xanthogranulomatous inflammation, abcess formation in the same fallopian tube. This is the first reported case, both lesions in the same patient. Immunesupression is the common etiologic factor for both lesions. Uncontrolled diabetes mellitus resulting in impaired leukocyte function and immunocompromised status may be predisposing factors. Preoperative diagnosis of both adenomatoid tumor and xantogranulamatous inflammation may be important in avoiding aggressive surgical intervention but mostly the exact diagnosis can only be made by pathologic examination.

Keywords: adenomatoid tumor, xanthogranulomatous inflammation, diabetes mellitus

INTRODUCTION

Adenomatoid tumors of the genital tract are rare and benign neoplasms and commonly seen in the reproductive period (1, 2), mostly in the uterus in the women and epididymis in the men (3-5). The incidence of adenomatoid tumours is % 0.1-0.2 (6, 7).

Xanthogranulomatous inflammation is an uncommon form of chronic inflammation that may affect various organs like kidney, gallbladder, stomach, bone, urinary bladder, testis, epididymis, vagina and endometrium (8). But xanthogranulomatous inflammation of the female genital tract, especially of the ovary is very rare (9, 10). This case report is to document xanthogranulomatous inflammation of the ovary with the uterine adenomatoid tumour in a patient with diabetes mellitus.

CASE REPORT

A 47 year old postmenopausal patient with one children was admitted to our gynecology department for routine control. During gynecological examination, there was a painless palpable mass in the right adnexial region and there was no pain during cervical movements. Ultrasonographic examination revealed a normal sized uterus and 72*42 mm cystic lesion with heterogenious ecogenity in the right adnexial region and there was a 120*40 mm sized collection of intraabdominal fluid with linear saptations in the douglas and bilateral paraovarian regions. She had intrauterine device for 2 years and she had it removed 3 years ago. The patient had diabetes mellitus for 6 years and she was using oral antidiabetics. She had neurogenic bladder due to diabetes mellitus for 3 years and she was doing self intermittant cateterization. She had a history of smoking for 6 years. Some of the laboratory parameters were as follows; hemoglobulin:9.8 g/dl, white blood count: 11.3 K/UL, glucose: 305 mg/dL, HbA1c: 12.5%, Ca 125: 41 IU/L, Ca 19-9: 42 kU/L, Ca 15-3: 20 kU/L, CEA: 2.6 ng/ml. Cervikovaginal smear was negative for intraepithelial lesions. Laparatomy was

[1] *Obstetrics and Gynecology Department, Yıldırım Beyazıt University, Ankara, Turkey*
[2] *Obstetrics and Gynecology Department, Ankara Atatürk Education and Research Hospital, Ankara, Turkey.*
[3] *Patology Department, Ankara Atatürk Education and Research Hospital, Ankara, Turkey*

Correspondence: *Özlem Şengül*
Ankara Atatürk Education and Research Hospital, Bilkent, Ankara, Turkey
Phone: +905056342770

E-mail: ozlem.sengul@yahoo.com

Received: 01 March 2017, Accepted: 21 Sep 2017

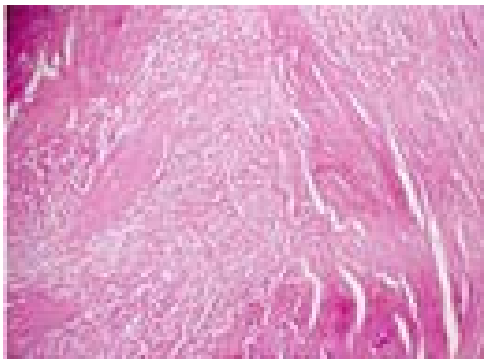

Figure 1: *Adenomatoid tumor of the uterus*

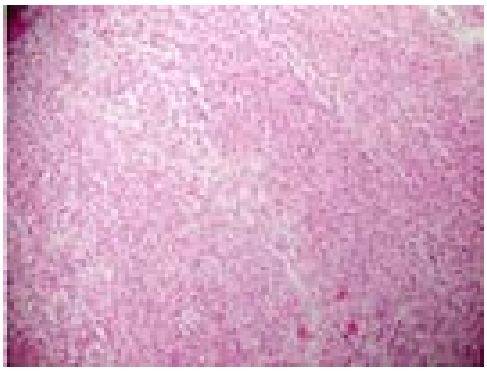

Figure 2: *Xanthoma cells in the ovary*

planned for the adnexial mass. Informed consent covering permission for the documentation of the case and for the operation was taken from the patient.

The patient was prepared for the operation and laparotomy was performed under general anesthesia. During laparatomic examination, there was ascites in the abdomen and a sample was taken for cytologic examination. There was a 10 cm cystic lesion in the right adnexial region with slight adhesions around. During the dissection of the cyst from the adhesions, 10 ml of purulent content was expelled. Total abdominal hysterectomy and bilateral salphingooferectomy were performed. The patient was given antibiotic treatment after the operation. The definitive pathology report came out as negative cytology for malignancy in the ascites, adenomatoid tumor of the uterus (14 mm in diameter) (**Figure 1**), right ovarian xanthogranulomatous inflammation (**Figure 2**), abcess formation in the right fallopian tube. The patient was discharged from the hospital on the second day of the operation and the recovery period was uneventful.

DISCUSSION

Adenomatoid tumors are benign mesothelial tumors (1, 2, 5). Tubular formation, solid growth or cystic areas may be seen in their histological pattern (11). The patients with adenomatoid tumors are usually asymptomatic or may have mild symptoms and a palpable mass (12). The preoperative differential diagnosis is usually difficult and the exact diagnosis is usually done by pathologic examination (6, 7, 13). It was suggested that magnetic resonance imaging may differentiate uterine adenomyoma from leiomyoma preoperatively (14). Manjunath et al. suggested that fine needle aspiration of the adenomatoid tumors may help to exclude malignancy and to avoid unnecessary aggressive surgical intervention (12). There was no recurrence, malign transformation or matastasis reported in the follow-up of the patients with adenomatoid tumors. It was also suggested that it may be related to immunosupresive conditions like hapatitis C (15) or renal tranplant (16, 17). Our patient was also asymptomatic and the diagnosis of adenomatoid tumor was done by the pathologic examination.

Xanthogranulomatous inflammation is one of the destructive forms of chronic inflammmation (8). Massive infiltration of the tissues by lipid-laden histiocytes mixed with lymphocytes, plasma cells and polymorphonuclear leukocytes are the histologic characteristics of the xanthogranulomatous inflammation (8). It may mimic pelvis tumors especially the malignant ones clinically and radiologically (18). Although the etiology is unclear, it is suggested that it may be related to endometriosis, infections, ineffective and inappropriate antibiyotic theraphy, systemic and chronic inflammatory diseases (8, 19). It was suggested that xanthogranulomatous inflammation may result from diminution of macrophagocytic function under the influence of systemic illness or corticosteroid excess (20). Uncontrolled diabetes

mellitus may result in impaired leukocyte function and infection leading to the xanthogranulomatous inflammation (21). This patient had also diabetes mellitus and history of intrauterine device usage as the predisposing factors. Right tubal abcess was also together with the xanthogranulomatous inflammation of the ovary probably showing the effect of infection which may also be a predisposing factor.

Both adenomatoid tumor of the uterus and xanthogranulomatous inflammation of the ovary are very rare lesions and they are seen together in this patient. This is the first reported case of both lesions in the same patient. Immunesupression is the common etiologic factor for both of them. Uncontrolled diabetes mellitus resulting in impaired leukocyte function and immunocompromised status may be the predisposing factors in this case. Both diseases should be come into mind especially in patients with chronic diseases and with a tendency to immunesupression. Preoperative diagnosis of both adenomatoid tumor and xanthogranulomatous inflammation may be important in avoiding aggressive surgical intervention but mostly this is not the condition and the exact diagnosis can only be made by pathologic examination.

REFERENCES

1. Nogales FF, Isaac MA, Hardisson D, et al. Adenomatoid tumors of the uterus: an analysis of 60 cases. Int J Gynecol Pathol. 2002;21(1):34–40.
2. Christensen C, Bichel P. Adenomatoidtumour of uterus. Case report. Br J ObstetGynaecol 1988;95:524–526.
3. Young RH, Silva EG, Scully RE. Ovarian and juxtaovarian adenomatoid tumor: a report of six cases. Int J Gynecol Pathol. 1991;10: 364-71.
4. Hes O, Pérez-Montiel DM, Alvarado Cabrero I, et al. Thread-like bridging strands: a morphologic feature present in all adenomatoid tumors. Ann DiagnPathol. 2003;7:273–277.
5. Tiltman AJ. Adenomatoidtumours of the uterus. Histopathology. 1980;4:437–43.
6. Livingstone EG, Guis MS, Pearl ML, Stern JL, Brescia RJ. Diffuse adenomatoid tumor of the uterus with a serosal papillary cystic component. Brief communication. Int J GynecolPathol. 1992;11(4): 288-92.
7. Klintorp S, Grinsted L, Franzmann MB. Adenomatoid tumor of the uterus. Eur J Obstet Gynecol Reprod Biol. 1993;50:255-7.
8. Zhang XS, Dong HY, Zhang LL, Desouki MM, Zhao C. Xanthogranulomatous Inflammation of the Female Genital Tract: Report of Three Cases. J Cancer. 2012;3:100–6.
9. Ladefoged C, Lorentzen M. Xanthogranulomatous inflammation of the female genital tract. Histopathology. 1988;13:541-51.
10. Pace EH, Voet RL, Melancon JT. Xanthogranulomatousoophoritis- an inflammatory pseudotumor of the ovary. Int J Gynecol Pathol. 1984;3:398-402.
11. Rappa F, Ternullo MP. Adenomatoid tumor. Patologica. 2006:98(2):164-6.
12. Manjunath GV, Nandini NM, Sunila. Fine needle aspiration cytology of adenomatoid tumour--a case report with review of literature. Indian J Pathol Microbiol. 2005;48(4):503-4.
13. Esin Y, Rukiye C, Gülhan A. Adenomatoid tumors of the uterus: morphological, histochemical and immunohistochemical findings in two cases. Türkiye ekopatoloji dergisi. 1999;5(1-2);48-52.
14. Mitsumori A, Morimoto M, Matsubara S, Yamamoto M, Akamatsu N, Hiraki Y. MR appearance of adenomatoid tumor of the uterus. J Comput Assist Tomogr. 2000;24(4):610-3.
15. Acıkalın MF, Tanir HM, Ozalp S, Dundar E, Ciftci E, Ozalp E. Diffuse uterine adenomatoid tumor in a patient with chronic hepatitis C virus infection. Int J Gynecol Cancer. 2009;19(2):242-4.
16. Duval H, Rioux-Leclercq N, Bauville E, Al Jaradi M, Burtin F. [Multinodular-adenomatoid tumor of the uterus in a patient with a renal allograft]. Ann pathol; 2008;28(4):308-10.
17. Tiras B.M, Noyan V, Süer O, Bali M, Edali N, Yildirim M. Eur J Obstet Gynecol Reprod Biol. 2000 Oct;92(2):205-7.
18. Kunakemakorn P, Ontai H. Pelvic inflammatory pseudotumor: A case report. Am J Obstet Gynecol. 1976;26:286–7.

19. Shalev E, Zuckerman H, Rizescu I. Pelvic inflammatory pseudotumor (Xanthogranuloma) Acta Obstet Gynecol Scand. 1982;61:285–6.
20. Chou SC, Wang JS, Tseng HH. Malacoplakia of the ovary, fallopian tube and uterus: a case associated with diabetes mellitus. Pathol Int. 2002;52(12):789-93.

◇◈◈◈◈◈◇

http://www.ejgm.co.uk