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Histiocytic endometritis

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 72
Final Diagnosis: Histiocytic endometritis
Symptoms: Vaginal discharge • fever • weakness
Medication: —
Clinical Procedure: Endometrial and cervical biopsy
Specialty: Pathology • Gynaecology

Objective: Rare disease

Background: Histiocytic or xanthogranulomatous endometritis, characterized by disappearance of endometrial mucosa and its replacement by sheets of lipid containing histiocytic cells, is very rare. Extensive internet and PubMed searches revealed only 19 cases reported to date. The pathogenesis of histiocytic endometritis seems to be inflammation due to post-menopausal cervical stenosis or as the result of cervical carcinoma. Histiocytic endometritis can infiltrate the myometrium and can mimic a malignancy.

Case Report: We report the case of a 78-year-old post-menopausal female with symptoms of vaginal discharge, fever, and weakness. Radiological investigation showed a mass lesion in the cervix, extending into the myometrium, suggestive of cervical carcinoma. The lesion was biopsied and histopathological examination led to the diagnosis of histiocytic endometritis with no evidence of malignancy.

Conclusions: Histiocytic endometritis, an inflammatory pathology, can mimic malignancy clinically as well as radiologically. Histopathological examination with extensive sampling of tissue is essential because presence of endometritis does not rule out malignancy.

Key words: histiocytic endometritis • xanthogranulomatous endometritis • cervical carcinoma

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Background

Histiocytic or xanthogranulomatous endometritis is a process seen following conditions like hematometra or pyometra, characterized by disappearance of endometrial mucosa and its replacement by sheets of lipid containing histiocytic cells [1]. Other variants are *nodular histiocytic hyperplasia* and *ceroid-containing histiocytic granuloma*. Histiocytic inflammation is an unusual condition that involves organs such as the kidney and gall bladder, which are subject to chronic obstruction with subsequent infection, but involvement of the uterus is rare [2]. Greater awareness of this condition is needed because it can mimic malignancy with infiltration into the myometrium. We present a case of histiocytic endometritis, clinically and radiologically diagnosed as cervical carcinoma with involvement of the uterine corpus.

Case Report

A 78-year-old woman, postmenopausal for the last 30 years, presented to our gynecology outpatient department with symptoms of vaginal discharge for the last 3 weeks, associated with weakness and fever. The discharge was foul smelling and yellowish with a reddish tinge. There was no history of urinary frequency, diabetes, or tuberculosis. Her parity score was P4L4, with all 4 pregnancies being full-term vaginal delivery and the last pregnancy was 50 years ago. The patient underwent tubal ligation 46 years ago and also gave a history of surgery for intertrochanteric fracture of the femur neck, which was uneventful.

On examination, the patient was moderately built and moderately nourished, with mild pallor. The abdomen was soft and non-tender. Per-speculum examination revealed the cervix, which was bleeding on touch. On vaginal examination, 6–8 weeks anteverted uterus with parametrial tenderness was noted.

Lab test revealed 11.3 gm% hemoglobin with 11800/cumm total leucocyte count, with other parameters being normal. Radiological examination was advised and the MRI result was reported as a mass lesion in the cervix, with irregular margins extending into two-thirds of the myometrium superiorly and the upper one-third of the vagina inferiorly, with loss of fat planes between the uterus and posterior wall of the bladder and anterior wall of the rectum (Figure 1).

In view of clinical and radiological findings, a preoperative diagnosis of cervical carcinoma was made. Cervical and endometrial biopsies were taken under anesthesia and sent for histopathological examination. Intra-operatively, 15cc of pus was drained from the endometrial cavity and sent for microbiological examination, which was found to be positive for gram-positive cocci.

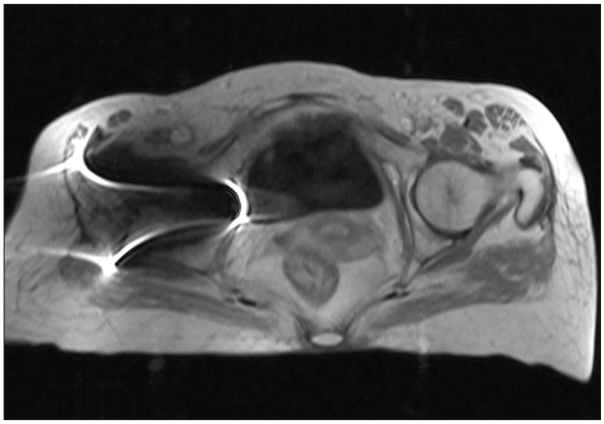

Figure 1. MRI showing uterine lesion with loss of fat planes between uterus and rectum.

Microscopic sections obtained from the tissue received for histopathological examination showed endometrial tissue with few glands and sheets of lipid containing foamy histiocytes in the stroma, along with diffuse infiltration by inflammatory cells (lymphocytes and plasma cells). A few areas of histiocytic aggregates were seen infiltrating the myometrium (Figures 2–5). Ziehl-Neelsen, PAS, and GMS staining revealed no specific organism.

Cervical biopsy revealed acute inflammation with regenerative atypia in the squamous epithelium. However, extensive study of endometrial and endocervical tissue revealed no evidence of malignancy.

Discussion

Although xanthogranulomatous inflammation is a distinguishing histopathological entity affecting various organs (chiefly the kidney and gall bladder), xanthogranulomatous endometritis is very uncommon [3–6]. The first case of XGE was described by Barua et al. [6] in 1978. Subsequent authors have added a few cases (Table 1): Buckley and Fox [7] in 1980 (2 cases), Ashkenazy et al. [5] in 1983 (4 cases), and Pounder and Iyer [3] in 1985 (1 case). Blanco et al. [8] in 1989 (1 case), Russack and Lammers [4] in 1990 (6 cases), Rivas and Phillipe [9] in 1996 (2 cases), and Kim and Lee [10] in 2002 described cases of nodular histocytic hyperplasia, which is considered to be a variant of XGE.

In 2006, Noack et al. [11] reported 1 case of XGE with lethal outcome. In 2007 Dogan-Ekici and Usubutun et al. [2] reported 1 case of XGE mimicking endometrial carcinoma, as in the present case, which was mimicking cervical carcinoma both clinically and radiologically.

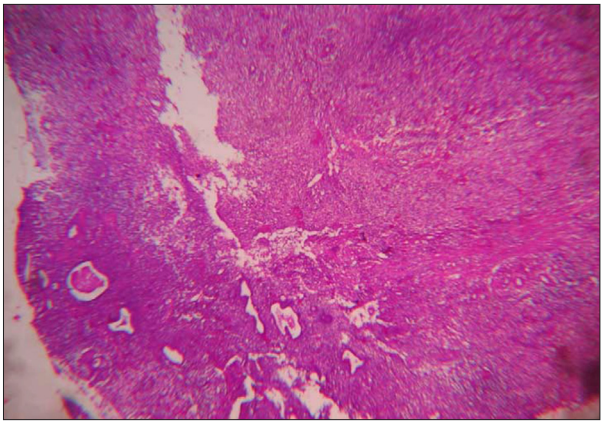

Figure 2. Microphotograph showing replacement of endometrial glands with histiocytic sheets with no evidence of malignancy (H&E: 4×).

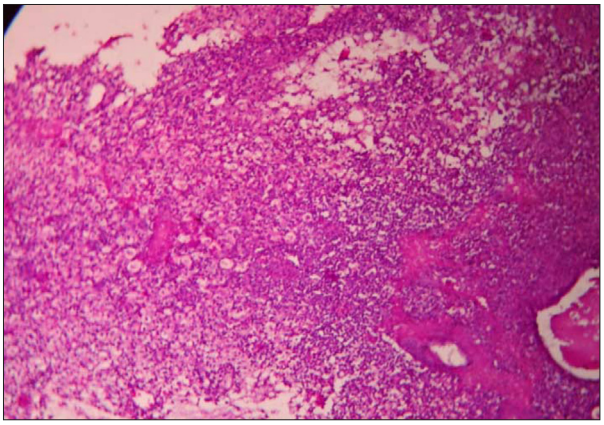

Figure 3. Microphotograph showing sheets of histiocytes with endometrial glands (H&E: 10×).

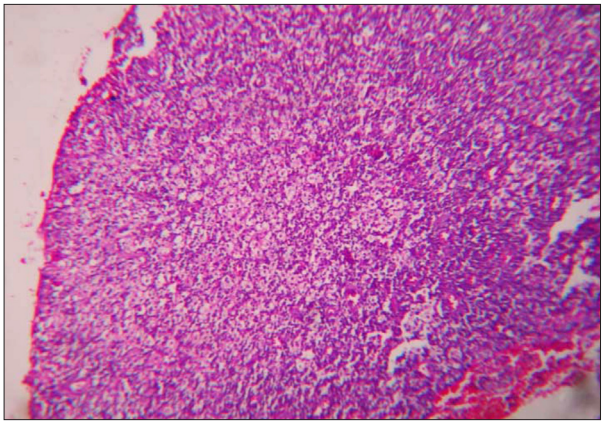

Figure 4. Microphotograph showing sheets of histiocytes (H&E: 10×).

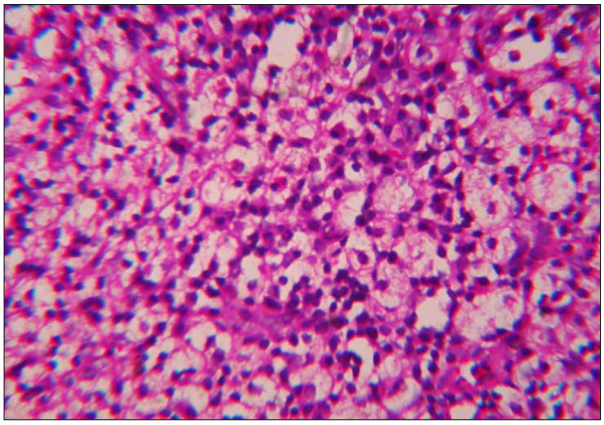

Figure 5. Microphotograph showing histiocytes with abundant foamy cytoplasm (H&E: 40×).

The term pseudoxanthoma and xanthogranuloma have also been used, but 'histiocytic endometritis' is preferred, which was coined by Buckley and Fox in 1980. Malakoplakia is another rare variant of histiocytic endometritis, which in addition shows Michaelis-Gutmann bodies [12].

The pathogenesis of histiocytic endometritis seems to be inflammation associated with pyometra due to post-menopausal cervical stenosis or as the result of cervical carcinoma. Many of the reported cases were associated with endometrial carcinoma [3–5]; in our case obstruction may have occurred because of post-menopausal cervical stenosis, as no evidence of malignancy was found on histopathology sections. Barua et al. [6] proposed infection (*E. coli* or *P. vulgaris*) as a cause of XGE. Our case also showed positivity for a microorganism (gram-positive cocci). However, Buckley et al. [7] and Dogan et al. [2] found no microorganism.

Because the histiocytes infiltrate the deeper tissues and can mimic malignancies such as clear-cell carcinoma [2], knowledge

Table 1. Distribution of reported cases.

Year	Authors	Number of case(s)
1978	Barua et.al.	1
1980	Buckley and Fox	2
1983	Ashkenazy et.al.	4
1985	Pounder and Iyer	1
1989	Blanco et.al.	1
1990	Russack and Lammers	6
1996	Rivas and Phillipe	2
2006	Noack et.al.	1
2007	Dogan-Ekici and Usubutun et.al.	1
Total		**19**

about the entity is essential for the pathologist, as it has an implication for treatment — histiocytic endometritis will require the correction of the cause, while malignancy will need surgery or chemotherapy.

However, presence of histiocytic endometritis does not rule out malignancy and requires extensive sampling of tissue to rule out any foci of neoplastic growth such as endometrial adenocarcinoma or cervical carcinoma, with which histiocytic endometritis has been found to be associated.

Conclusions

Histiocytic endometritis can mimic malignancy clinically as well as on imaging studies, as seen in our case. Pathologists should be aware of the entity and should rule out malignancy by extensive tissue sampling and histopathological examination.

Acknowledgement

To Departement of Radiodiagnosis, KIMSU, Karad.

References:

1. Rosai and Ackerman, Ch: Female reproductive system-Uterus Corpus. In. Surgical Pathology, 10th ed, Elsevier, 2012; 2: 1482
2. Dogan-Ekici A, Usubutun A, Kucukali T Ayhan A: Xanthogranulomatous Endometritis: A Challenging Imitator of Endometrial Carcinoma. Infect Dis Obstet Gynecol, 2007; 2007: 34763
3. Pounder DJ, Iyer PV: Xanthogranulomatous endometritis associated with endometrial carcinoma. Arch Pathol Lab Med, 1985; 109(1): 73–75
4. Russack V, Lammers RJ: Xanthogranulomatous endometritis. Report of six cases and a proposed mechanism of development. Arch Pathol Lab Med, 1990; 114(9): 929–32
5. Ashkenazy M, Lancet M, Borenstein R, Czernobilsky B: Endometrial foam cells. Non-estrogenic and estrogenic. Acta Obstet Gynecol Scand, 1983; 62(3): 193–97
6. Barua R, Krikland JA, Petrucco OM: Xanthogranulomatous endometritis: case report. Pathology, 1978; 10(2): 161–64
7. Buckley CH, Fox H: Histiocytic endometritis. Histopathology, 1980; 4(1): 105–10
8. Blanco C, Fernandez F, Buelta L et al: Xanthomatous endometritis. Applied Pathology, 1989; 7(4): 273–76
9. Rivasi F, Philippe E: Histiocytic endometritis. Report of two cases. Ann Pathol, 1996; 16(2): 112–14
10. Kim KR, Lee YH, Ro JY: Nodular Histiocytic Hyperplasia of the endometrium. Int J Gynecol Pathol, 2002; 21(2): 141–46
11. Noack F, Briese J, Stellmacher F et al: Lethal outcome in xanthogranulomatous endometri tis. APMIS, 2006; 114(5): 386–88
12. Robboy's Ch: Endometritis, metaplasias, polyps and miscellaneous changes. In. Pathology of the female reproductive tract, Elsevier, 2nd ed, 2009; 347–48