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Malakoplakia of unilateral fallopian tube, a rare entity at an unusual site: a case report

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ABSTRACT

Malakoplakia is an uncommon inflammatory condition usually affecting the genitourinary tract characterized by an infiltrate of large granular eosinophilic macrophages containing unique concentrically laminated siderocalcific structures called Michaelis-Gutmann bodies. It has been associated with infectious, tumors and immunocompromised states.
A 40-year-old woman, mother of 7 children presented with pain abdomen, polymenorrhagia and fever for 4 days duration. Total abdominal hysterectomy was performed. Specimen of uterus with cervix with bilateral adnexae was received. Left fallopian tube showed hydrosalpinx grossly. Section studied from the tube showed showed infiltration of the lamina propria by histiocytes with lymphocytes. The histiocytes (von Hansemann cells) had dense eosinophilic cytoplasm, vesicular nuclei and micronucleoli . There were round, basophilic intracellular inclusions (Michaelis-Gutmann bodies) of varying sizes, many calcified. Most of these inclusions were positive with the periodic acid-Schiff stain and focally for Perls' Prussian blue stain. The muscularis and serosa layer also showed chronic inflammatory infiltrate. Malakoplakia at all sites share the same histological features, with the presence of Michaelis-Gutmann bodies being pathognomic. It can clinically simulate tumours and can be associated with tumours, infections and immunosuppression. Inefficient killing of the microorganism by the macrophages underpins the pathogenesis. Malakoplakia of female genital tract is a rare entity. To the best of our knowledge, we are presenting the third reported case of malakoplakia of fallopian tube in the literature.

INTRODUCTION

Malakoplakia is an inflammatory reaction to organisms, which include bacteria, mycobacteria, fungi and occasionally parasites. It can simulate tumours and may result in diagnostic difficulties. It was initially described in the early 1900s as soft yellowish plaques found on the mucosa of the urinary bladder. Usually occurs in the urinary tract, it has been described in almost all organs [1]. It is a rare inflammatory condition which makes its presence known as a papule, plaque or ulceration that usually affects the genitourinary tract. However, it may also be associated with other bodily organs. Microscopically it is characterized by the presence of foamy histiocytes with basophilic inclusions called Michaelis-Gutmann bodies. It usually involves gram negative bacteria. It is thought to result from the insufficient killing of bacteria by macrophages. Therefore, the partially digested bacteria accumulate in macrophages known as Von-Hansemann cells and leads to a deposition of iron and calcium [2]

The impairment of bactericidal activity manifests itself as the formation of an ulcer, plaque or papule. It is associated with patients with a history of immunosuppression due to lymphoma, diabetes mellitus, renal transplantation, or because of long-term therapy with systemic corticosteroids. We describe a case of malakoplakia of unilateral fallopian tube, which to the best of our knowledge is the third overall case reported in the literature.

CASE REPORT

A 40-year-old woman, mother of 7 children presented with pain abdomen, polymenorrhagia, generalized weakness and fever for 4 days duration. On examination, abdomen was soft, tender

1a

1b

Fig 1a & 1b : Cut section of fallopian tube revealing yellow plaque.

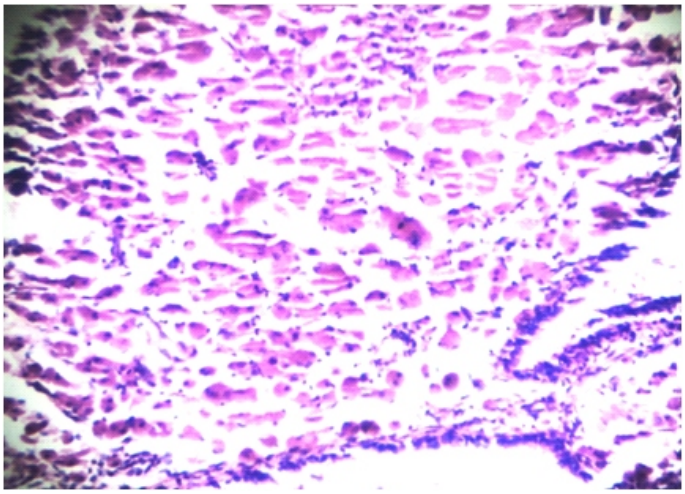


Fig 2. Lamina propria showing histiocytes and lymphocytes (100X H&E)

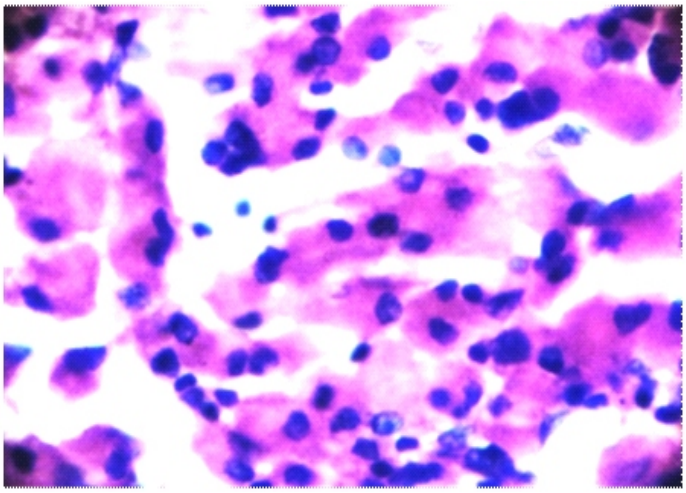


Fig 3. Von-Hansemann cells with Michaelis-Guttman bodies (400X H&E)

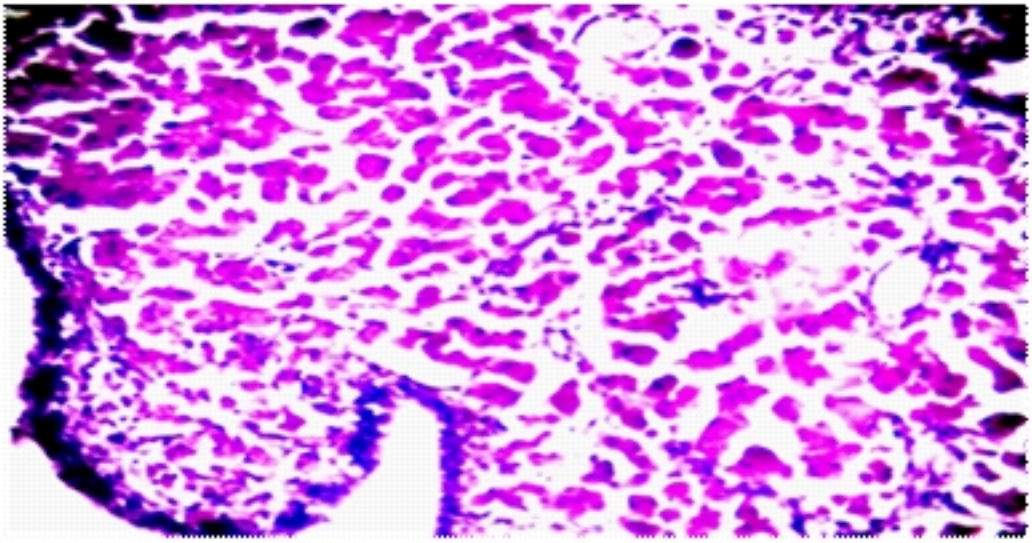
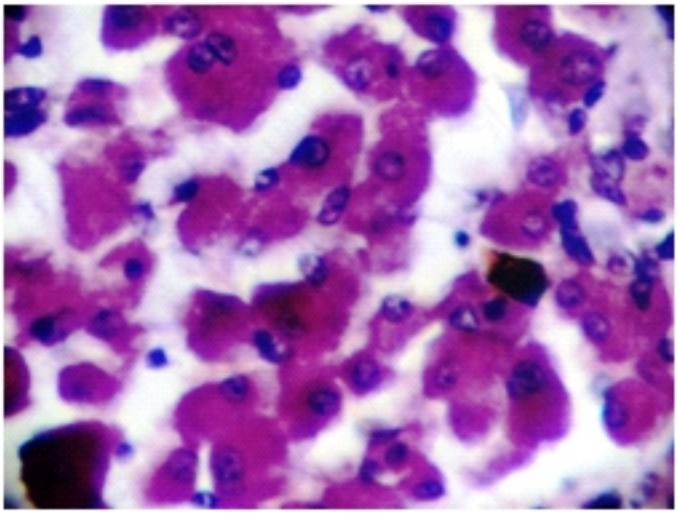


Fig 4. Lamina propria containing histiocytes showing PAS positivity (100X)



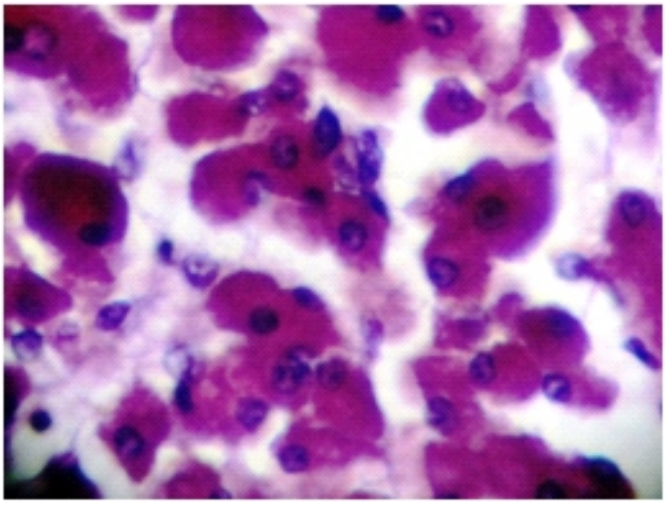


Fig 5a &5b : Vonhansemann cells showing " targetoid appearance" and "laminations" (400X PAS)

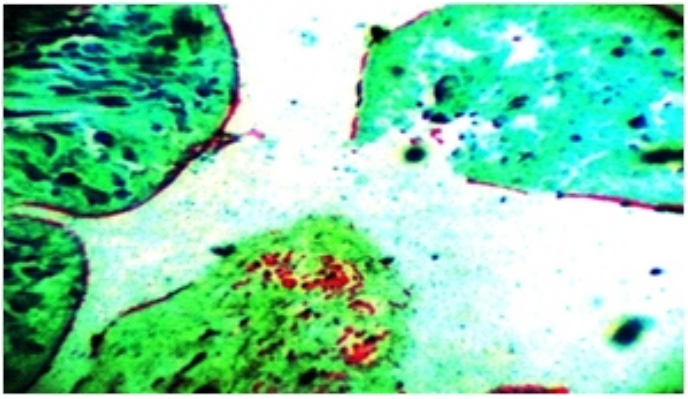


Fig 6 : Histiocytes showing Perl's Prussian blue stain positivity (100X)

and an incisional hernia was present. Per speculum examination revealed hypertrophy of cervix and uterus was bulky and tender. Hematological examinations revealed mild leukocytosis, other radiological examinations and systemic examinations were within normal limits. A preliminary diagnosis of Chronic Pelvic inflammatory disease was made due to bacterial infection. A Total abdominal Hysterectomy was performed.

Pathological findings:

The specimen consisted of a uterus with attached cervix and bilateral fallopian tubes and ovaries weighing 180 gm. The uterus measured 10x7x5cm. The right ovary measured 3x2x1cm and the right fallopian tube measuring 5cm. The left ovary measured 3.5x2.5x1 cm and the left fallopian tube showed hydrosalpinx measuring 6x4x2.5 cm. On cut section 4-5 ml of clear cystic fluid drained out with yellowish plaques noted (Fig.1a & 1b). Sections studied through cervix showed features of chronic cervicitis. Sections through endometrium showed secretory phase and myometrium was unremarkable. Sections studied through bilateral ovaries and right fallopian tube showed normal histology. Histopathologic examination of the left fallopian showed infiltration of the lamina propria by histiocytes and lymphocytes (Fig.2). The histiocytes (von Hansemann cells) had dense eosinophilic cytoplasm, vesicular nuclei and micronucleoli (Fig.3).There were round, basophilic intracellular inclusions (Michaelis-Gutmann bodies) of varying sizes, many calcified .Some were laminated while others appeared homogenous. Most of these inclusions were positive with the periodic acid-Schiff stain (Fig 4) and had a dense central core with a targetoid appearance (Fig 5a & 5b).These were focally positive for Perls' Prussian blue stain (Fig.6). The muscularis and serosa layer also showed chronic inflammatory infiltrate, plasma cells, and occasional neutrophils.

DISCUSSION

The first human case of malakoplakia was described by Von Hansemann, who coined the term 'malakoplakia' [3]. The aetiology of this condition is probably inflammatory. This benign non-neoplastic condition is believed to result from inadequate killing of bacteria, most commonly *Escherichia coli,* by a defect in monocytes and macrophages phagolysosomal activity. This response can also be seen with mycobacterial and fungal infections in immunocompromised patients. More than one organ can be affected simultaneously. Clinically and macroscopically malakoplakia can simulate tumours or abscesses. Histologically sheets and aggregates of histiocytes (von Hansemann cells) with fine eosinophilic granular cytoplasm are seen on haematoxylin and eosin stain. They are characterised by intracellular and extracellular, round basophilic concretions, called the Michaelis-Gutmann (MG) bodies. These stain with PAS diastase, von Kossa stain and Perls' Prussian blue. Electron microscopically, MG bodies show concentric crystalline laminations with a dense central zone containing partially digested bacteria and a thin outer zone. Immunohistochemically the cells are positive with CD68 and lysozyme. Earlier Aikat *et al* and Chou *et al* have reported cases of malakoplakia in fallopian tube in 1973 and 2002 respectively[4,5].

The exact aetiology of malakoplakia is ill understood. Malakoplakia is diagnosed primarily on histological grounds. Irrespective of the site, all malakoplakias share the same morphological features. Gram-negative bacteria, most commonly *E. coli*, have been frequently isolated from the cases of genitourinary malakoplakia. Various organisms that have been associated with this condition include *E. coli*, *Mycobacterium tuberculosis, Shigella boydii, Paracoccidioides species, Rhodococcus equi, Yersinia enterocolitica, Klebsiella pneumoniae, Proteus mirabilis, Staphylococcus aureus, Pseudomonas aeruginosa,* Enterobacter aerogenes and Taenia species [6]. The initial event in the pathogenesis of malakoplakia is partial digestion of the offending organism in macrophages by

phagolysosomes. These are eventually damaged, resulting in calcification. Some studies suggest that the underlying defect is a 3', 5'-guanosine monophosphate dehydrogenase deficiency, causing diminished phagolysosomal and bactericidal activity [7,8]. Malakoplakia of fallopian tube is a very rare entity and hence we are presenting this interesting case report.

CONCLUSION

Malakoplakia at all sites share the same histological features, with the presence of Michaelis-Gutmann bodies being pathognomic. It can clinically simulate tumours and can be associated with tumours, infections and immunosuppression. Inefficient killing of the microorganism by the macrophages underpins the pathogenesis.It is a chronic inflammatory process which most often affects the urinary tract. Malakaoplakia of fallopian tube is a very rare entity because of its unusual location. Further investigations should be carried out to detect the underlying cause.

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