Coexistence of malacoplakia and mullerianosis in the urinary bladder: An uncommon pathology

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ABSTRACT
Malacoplakia is an inflammatory lesion which can affect any organ in the body but predominantly affects the genitourinary system and mainly the bladder. Malacoplakia of the bladder has variable presentations and is associated with urinary infection or immunosuppression. Mullerianosis of the bladder is a rare lesion that consists of two out of the three tissues, endometriosis, endocervicosis, or endosalpingiosis. It is usually associated with a previous cesarean section or pelvic surgery. The diagnosis is confirmed on histopathological examination. Malacoplakia and mullerianosis are usually isolated lesions of the bladder. We present a unique case of coexistence of malacoplakia and mullerianosis in the urinary bladder, reported for the first time in the literature.

INTRODUCTION
Malacoplakia of the bladder is a rare lesion. It is characterized by granulomatous inflammatory reaction probably due to a defect in the intraphagolysosomal digestive activity of the macrophages and monocytes, leading to an inadequate killing of the ingested bacteria. Malacoplakia can be present in any organ, but is more commonly seen in the genitourinary system particularly the bladder. Mullerianosis is a lesion characterized by presence of two or more tissues of endometriosis, or endocervicosis containing endocervical glands, and tubular epithelium of endosalpingiosis. It was first described by Young and Clement in 1996[1] and is usually seen in fertile females with symptoms of hematuria or pain during menstruation. We present an extremely rare case of coexistence of these two lesions in the urinary bladder.

CASE REPORT
A 25-year-old female presented with dysuria and lower abdominal pain, especially during menstruation. She had a previous cesarean section. The physical examination was within normal limits. The sonography showed a 55 mm x 40 mm mass lesion in the bladder. Contrast computed tomography scan of the abdomen and pelvis showed an ill-defined heterogeneously enhancing solid-cystic lesion, with predominantly cystic component, over the antero-superior part of the urinary bladder, extending anteriorly in the prevesical space and infiltrating into the anterior abdominal wall muscles, in the region of the previous cesarean section scar. The lesion measured 50 mm x 29 mm x 26 mm [Figure 1a]. The lesion was not extending towards the umbilicus and the rest of the bladder was normal. Cystoscopy showed a mass over the dome of the bladder. The mucosa over the mass lesion was normal. As the mass was mimicking malignancy, decision was taken to excise it and send for histopathological examination. A lower abdominal exploration was performed with a Pfannenstiel incision including the cesarean scar. There was a mass lesion in the abdominal muscles on the right lateral side of the midline extending till the bladder dome. Complete excision of the mass with partial cystectomy

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was performed [Figure 1b]. Histopathological examination revealed the presence of plasma cells, giant cells with calcium deposits, foamy histiocytes, and presence of Michaelis–Gutmann bodies, which is pathognomonic of malacoplakia. Periodic acid–Schiff staining was performed to confirm the finding of Michaelis–Gutmann bodies [Figure 2a]. Further confirmation was obtained by the Von Kossa staining which showed the presence of calcium in the Michaelis–Gutmann bodies [Figure 2b]. All the above findings were suggestive of malacoplakia. In addition, histopathology also showed features of mullerianosis. It consisted of endometrial glands and surrounding endometrial stroma and also the glands that were lined by columnar epithelium [Figure 2c]. The presence of these two histological features confirmed the diagnosis of mullerianosis.

The patient had uneventful postoperative course. Her symptoms of dysuria during menses completely subsided after the surgery.

DISCUSSION

Malacoplakia and mullerianosis are benign rare lesions of the bladder and it is important to rule out malignancy on the histopathological examination after surgery. Malacoplakia is a rare lesion and the exact etiology is unknown, but it is commonly associated with chronic *Escherichia coli* infections and immunosuppression. It has characteristic histological features such as the presence of foamy histiocytes known as Van Hansemann cells in the inflammatory infiltrates, with the basophilic inclusions known as the Michaelis–Gutmann bodies.

Michaelis–Gutmann bodies are intracytoplasmic or extracellular oval basophilic structures of targetoid or bull’s eye, or concentric owl eye appearance consisting of calcium phosphate crystals with undigested bacterial components trapped in the lysosomes of macrophages and monocytes. The treatment options include antibiotics and surgical excision.[2] Malacoplakia can also present as a mass in the urachus, which is difficult to differentiate from malignant lesions.[3] In our case, the presence of malacoplakia on histopathological examination was a surprise as there were no clinical features to suggest this diagnosis.

Mullerianosis is a lesion which contains two or more Mullerian tissues, endometrial, cervical, or tubal, in the muscularis propria of the urinary bladder. It usually presents as a mass and can mimic a neoplastic lesion. The cause of mullerianosis is unclear, but it can occur after pelvic surgery or cesarean section. It can also affect patients without any previous surgery, where it is believed to be due to differentiation of the Mullerian epithelium into endometrial, endocervical, and tubal types.[4] The mass, in our case, was just under the cesarean scar in the abdominal muscles and was extending till the bladder, probably suggesting secondary to the previous cesarean section surgery. In our case, the histopathology showed endometrial glands and lining of columnar epithelium suggestive of tubal pathology. Mullerianosis can be confused with other lesions of the bladder such as cystitis cystica, cystitis glandularis, urachal remnants, and nephrogenic adenoma and should be ruled out with careful pathological examination.[5] In this case, though clinically it favored mullerianosis, due to previous history of cesarean section and dysuria during menstruation, the histopathological
finding was other way round. The features of malacoplakia were more predominant than mullerianosis.

CONCLUSION

Malacoplakia and mullerianosis can coexist in the bladder. A careful histopathological examination is essential to rule out such a rare bladder lesion with two different pathologies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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