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Southeast Asian Journal of Case Report and Review

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Case Report

Endosalpingiosis of urinary bladder presenting as a polypoidal mass: A case report and review of clinicopathological features

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Abstract

A 51 year of female presented to the outpatient department of a hospital with a complain of lower abdominal pain and heaviness for which an Ultrasonography of Kidney, Ureter and Bladder was advised which showed a hypoechoic cystic lesion (18.7x10.8mm) with mild vascularity in the indenting mid part of posterior wall of urinary bladder. The patient underwent Cystoscopy and Transabdominal excision of the Space occupying lesion. Histopathological study of the space occupying lesion demonstrated variably sized cystically dilated glands in the muscularis propria, lined by ciliated columnar tubal like epithelium. Immunohistochemically these glands were positive for PAX8. Accordingly, a diagnosis of endosalpingiosis of urinary bladder was made.

Keywords: Endosalpingiosis, Urinary bladder, Mullearianosis

Received: 20-01-2024; **Accepted:** 11-03-2025; **Available Online**: 28-04-2025

1. Introduction

Mullearianosis of the urinary bladder is an extremely rare benign condition, first described by Young and Clement in 1996.[1] Of which Endosalpingiosis is the rarest. The term 'endosalpingiosis' which was introduced by Sampson in 1930, described the local proliferative and invasive properties of tubal mucosa following surgical interruption.[2] Noval challenged this definition and noted these lesions were also seen in patients without a history of tubal surgery and who had a history of pelvic inflammatory disease.[3]

Endosalpingiosis of the urinary bladder is also an extremely rare condition characterized histopathologically by the sole presence of tubal type epithelium without other mullerian components in the lamina propria and muscularis propria of the urinary bladder.[4] Till date six cases has been described in literature, including the one mentioned in the article.[1]

In this case report, we describe a case of endosalpingiosis of the urinary bladder which presented as a polypoidal mass arising from urinary bladder wall and review the clinicopathological features of this entity.

2. Case Summary

A 51-year-old postmenopausal female came to the department of urology with complaint of lower abdominal pain. There was no complaint of fever, dysuria or hematuria. On investigating: routine examination of urine, complete blood counts, blood urea, and serum creatinine were within normal range. The urine culture showed no growth. The ultrasonography of kidney ureter bladder showed a hypoechoic cystic lesion (18.7 x 10.8 mm) with mild vascularity seen indenting mid part of the posterior wall of urinary bladder square (**Figure 1**). The urinary bladder was otherwise normal in size. There was no intra cystic space occupying lesion or calculus identified. The mucosa was normal. There was no diverticulum or ureterocele. Multi detected computerized tomography of whole abdomen showed 20 x 12 x 18 mm space occupying lesion in the lower posterior urinary bladder wall projecting

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DOI: https://10.18231/j.sajcrr.2025.004

intraluminally and is 19 mm supero-medial to right vesicoureteric junction which on adding contrast showed enhancement with tiny non-enhancing portion in the center. The perivesical fat planes were normal. The uterus and bilateral adnexa were normal in anatomy. (**Figure 2**).

Based on above radiological findings, a decision of cystoscopy, transurethral mucosal demarcation and incision around intravesical portion of urinary bladder space occupying lesion, trans abdominal laparoscopic partial cystectomy followed by bilateral Double J stenting was taken.

On cystoscopy a smooth round soft submucosal space occupying lesion was noted with prominent vascular marking (**Figure 3**). Partial cystectomy including the space occupying lesion and block with a 0.5 cm rim of normal bladder mucosal tissue was excised laparoscopically. Grossly the mass was cystic.

Histopathological study showed detrusor muscle and fibro adipose tissue interspersed by cystically dilated glands lined by benign ciliated columnar epithelium which was immunohistochemically positive for PAX-8 and ER, thereby emphasizing the mullerian origin (**Figure 4**). Due to absence of any other mullerian derived components, endometrial stroma and hemosiderin laden macrophages and presence of fallopian tube like epithelium a diagnosis of endosalpingiosis was made. Postoperatively the patient is doing well. On day 7 post- operative, the patient was discharged with catheter in situ. The post-operative x-ray showed bilateral Double J stent with no extravesical leakage.

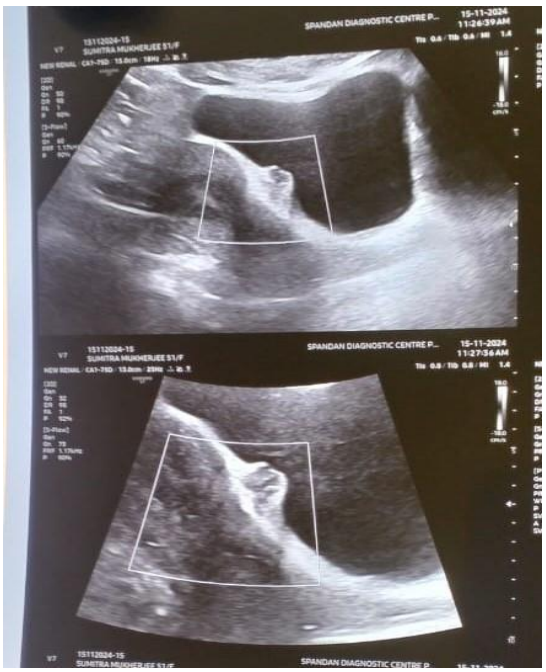

Figure 1: Ultrasonography of kidney ureter bladder showed a hypoechoic cystic lesion (18.7 x 10.8 mm) with mild vascularity seen indenting mid part of the posterior wall of urinary bladder square

Figure 2: Multi detected computerized tomography of whole abdomen showed 20 x 12 x 18 mm space occupying lesion in the lower posterior urinary bladder wall projecting intraluminally and is 19 mm supero-medial to right vesicoureteric junction which on adding contrast showed enhancement with tiny non-enhancing portion in the center.

Figure 3: Cystoscopy shows a smooth round soft submucosal space.

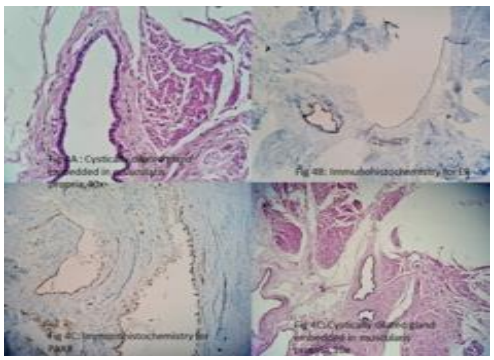

Figure 4: Entrapped glandular structures highlighted by ER and PAX8.

3. Discussion

Endosalpingiosis is the presence of ectopic cystic glands outside fallopian tube and are lined by same tube like epithelium. It may occur in pelvic organs like ovary, serosal aspect of fallopian tube, uterine serosa, myometrium or pelvic peritoneum and extra pelvic organs like urinary bladder, retroperitoneal lymph nodes or axillary lymph

nodes. Endosalpingiosis is a forgotten entity in close proximity to endometriosis. It is an important clinical entity since it may be very commonly associated with chronic pelvic pain. In our case the patient presented with lower abdominal pain.

Unfortunately, this diagnosis is often overlooked since it is mistaken for endometriosis and ablated with diathermy. Instead, if excision of suspicious lesions is done rather than ablating lesions, it may increase our knowledge of endosalpingiosis and may be the most suitable treatment for the condition.[5] In our case, since she presented with a submucosal space occupying lesion of urinary bladder, it was excised and delivered en bloc instead of ablation.

Endosalpingiosis of the urinary bladder in particular is an extremely rare condition, characterized histopathologically by the presence of tubal type epithelium without other mullerian components in the lamina propria and muscularis propriety of urinary bladder.[6,7] It was described as an isolated entity by Young and Clement in 1996.[8,9] Imaging appearance of the entity is of not much help, hence awareness of the condition is helpful in broadening the differential diagnosis of bladder mass. We report a case which presented as a diagnostic challenge in imaging studies. The differentials thought of on imaging were leiomyoma, rhabdomyoma, lipoma etc.

The pathogenesis of endosalpingiosis remains completely hypothetical. Some of the proposed hypothesis are as follows: The implantation theory by Young and Clement proposes that mullerian tissue can implant in the urinary bladder wall during caesarean section or pelvic surgery.[1] In our case there is no history of previous caesarean section. However, there is a history of laparoscopic cholecystectomy 15 years back.

Cystoscopic examination is next best investigation which can be both diagnostic and therapeutic. It can be used in management either to obtain a tissue sample or implement the treatment process. It may reveal a vascular lesion or mucosa covered polyp as was seen in our case also.

However, in order to arrive to a specific diagnosis, it is not possible without histopathological examination of tissue. Histopathology is compulsory for the diagnosis of endosalpingiosis.

On microscopy, although it is an obvious diagnosis, yet there are few mimics which need to be differentiated. The histopathological differential diagnostic considerations of Mullearianosis and Endosalpingiosis of the urinary bladder include several benign conditions, such as cystitis glandularis, urachal remnant, nephrogenic adenoma, and adenocarcinoma.[9] Cystitis glandularis which mostly arise from surface epithelium extending up to the lamina propria and the glands are lined by PAX-8 negative non-ciliated columnar or cuboidal epithelium. Nephrogenic adenoma/metaplasia is another differential which microscopically exists in papillary or tubular patterns and are lined by non-ciliated. Cuboidal to flattened epithelium. Microscopic urachal remnant it can also be considered as a differential which can be excluded since it is rare in adults and are lined by cuboidal to flattened urothelium with occasional intestinal metaplasia. Adenocarcinoma can also be considered as a histopathological differential diagnosis, due to region of the infiltration of the glands in the muscularis propria. However, it can be distinguished from endosalpingiosis by observing features of malignancy like atypia, nuclear pleomorphism and mitosis.

4. Conclusion

This study emphasizes the need for adequate and elaborate clinicopathological inputs while considering the differential diagnosis of and intramucosal /submucosal bladder lesion. It also highlights the significance of histopathological examination in establishing a definitive diagnosis.

5. Conflict of Interest

None.

6. Conflict of Interest

None.

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Cite this article: Sutradhar A, Pandit K. Endosalpingiosis of urinary bladder presenting as a polypoidal mass: A case report and review of clinic pathological features,*Southeast Asian J Case Rep Rev.* 2025;12(1):**13-15.**