Int J Clin Exp Pathol (2009) 2, 91-94
www.ijcep.com/IJCEP801001

Case Report

Endocervicosis of the Bladder: Report of a Case and Review of the Current Literature

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Received 18 Jan 2008; Accepted and available online 28 Jan 2008

Abstract: An important variety of metaplastic lesions of the urinary bladder have been reported in the medical literature up to date. Among those of Müllerian origin, endocervicosis is the most infrequent. We report a 67 years old woman who presented with a history of left flank pain and dysuria for the past 2 months. Imaging studies revealed a solid mass in the posterior bladder wall protruding into the lumen. Transurethral resection of the lesion was performed. Both pathologic examination and immunohistochemistry confirmed the diagnosis of endocervicosis. The patient was monitored with 6 months interval follow up and remains disease free 12 months post surgery.

Key Words: Bladder, endocervicosis, metaplasia, neoplasm

Introduction

The urothelium frequently undergoes metaplasia (changes in its morphology into another cell type which is considered aberrant for that location), presumably as a reaction to a local stimulus (e.g. chronic inflammation, urinary tract infection, calculi, diverticuli, catheterization, or surgical procedure). Genito-urinary involvement with one of the triad of non-neoplastic secondary Müllerian lesions (endocervicosis, endometriosis or endo-salpingiosis) occurs in 1-2% of cases. Most of these cases regard endometriosis [1]. On the contrary, endocervicosis is very rare. It was first described as a distinct entity by Clement and Young in 1992 as a benign variant of endometriosis [2]. Few reports of this entity concerning exclusively women of reproductive age have been described since.

Case Presentation

A 67 years old woman presented to the outpatient department of our hospital with a history of mild persistent left flank pain, nocturia, dysuria and frequency for the past 2 months. Her history was significant for 2 prior Caesarean sections at the age of 40 and 42, respectively. On physical examination, blood pressure was 135/80. Palpation of the lower left abdomen caused diffuse pain. Routine laboratory tests were normal. Urine culture and cytological analyses showed no malignancy. Transabdominal ultrasound revealed an immobile, 3.3 × 4.9 × 4 cm solid mass in the posterior bladder wall protruding into the lumen of the bladder. A mild dilatation of the left renal pelvis and upper left ureter was revealed also. Computed tomography

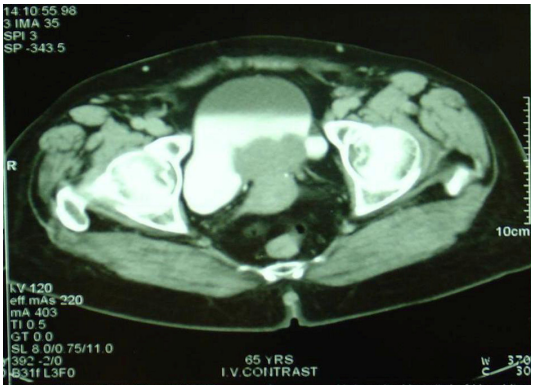

Figure 1 Computerized tomography imaging of the pelvis showing a solid mass arising from the posterior bladder wall.

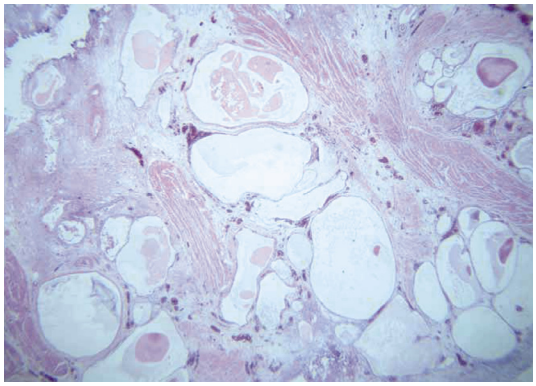

Figure 2 Histopathological image showing the mucinous glands between the muscular tissue of the bladder wall.

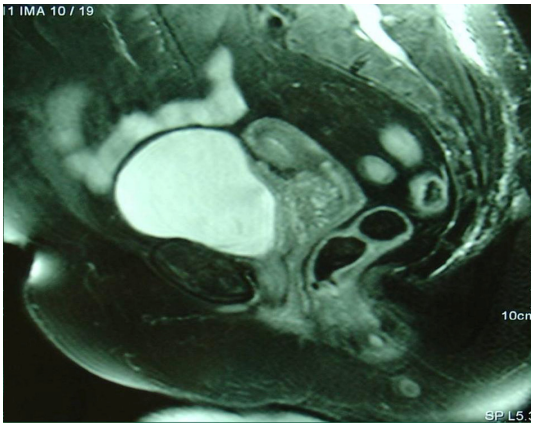

Figure 3 Magnetic resonance imaging revealing focal thickness of the bladder wall due to the development of fibrous tissue extending from the lower part of the external surface of the bladder to the anterior surface of the uterus.

imaging of the pelvis confirmed this finding (**Figure 1**).

A large submucosal lesion extending from the trigone to the left collateral wall and the posterior portion of the dome of the urinary bladder was seen on cystoscopy. An extensive transurethral resection of the tumour was performed while a ureteral stent was placed into the left ureter. A 3 way Foley catheter was placed in order to monitor the bladder bleeding. The pathologic examination of the biopsy material revealed extensive involvement of the bladder by mucinous glands that penetrated into the muscularis propria of the bladder wall. These glands were haphazardly arranged, and many had expanded into mucus-filled cysts of various sizes (**Figure 2**).

The lining of the cysts ranged from a single layer of columnar cells with abundant pale cytoplasm to ciliated cuboidal and flattened cells. There was no nuclear atypia or mitotic activity. There were focal areas that contained ruptured cysts, with extravasation of mucin into the muscularis propria. The stroma that surrounded these glands was edematous and contained a chronic inflammatory infiltrate. On immunohistochemistry, the epithelial elements of the lesion were positive for cytokeratin 7 and negative for cytokeratin 20. The majority of the glands displayed strong expression of estrogen receptors. The appearances were those of a non-neoplastic lesion, and the final diagnosis of endocervicosis was established.

Magnetic resonance imaging of the pelvis and magnetic urography performed after the displacement of the Foley catheter revealed a focal thickness of the bladder wall due to the development of fibrous tissue, a finding which was attributed to the recent history of endocervicosis (**Figure 3**).

Cytological analyses of uterus and vagina smear showed no malignancy. The patient was monitored with 6 months interval follow up, with transabdominal ultrasound, cystoscopy and endoscopically guided biopsies of the bladder. The patient remains disease free at 12 months interval.

Discussion

Endocervicosis of the urinary bladder is a very rare benign condition characterized by mucinous endocervical epithelium within the detrusor muscle of the bladder [3]. It is believed to be a lesion of Müllerian origin, arising from Müllerian rests after their differentiation to intracervical tissue [4]. This hypothesis has been supported by the following lines of evidence: main occurrence in women of reproductive age, location in the posterior bladder wall and coexistence with endometriosis and endosalpingiosis [5, 6]. When immunohistochemical phenotype of a case of endocervicosis was compared with four normal uterine endocervices, endocervicosis glands displayed stronger expression of uterus reactive antibodies such as HBME-1, estrogen receptor (ER) and progesterone receptors (PR) [7]. These

findings confirmed the endocervical nature of endocervicosis and constituted further arguments for the Müllerian origin hypothesis. The aetiology and pathogenesis of this entity remain unknown. There is evidence to suggest that it is hormone and age-related. However, although it is thought to be seen in women of child bearing age [8-10] in our case it has been diagnosed in a post menopausal woman; the occurrence in post menopausal women is intriguing and possibly disconnects its aetiology and pathogenesis from the hormonal mechanisms.

Among several predisposing factors proposed to have an association with the development of endocervicosis, history of previous Caesarean section seems to be the most questionable [11]. It is however interesting that in our case, as in all previous reports, a background of Caesarean sections was present [1-12]. On the other hand, since the proliferative index of endocervicosis cells was found to be within the normal range established for endocervical glands [7], it is plausible that there is no evidence of a distinct proliferative aetiopathogenetical background. Therefore, a background of uterine endocervical migration during previous surgery cannot be excluded.

To the best of our knowledge, there are less than twenty five cases reported so far. Although it is usually an incidental histologic finding, it may cause non-specific symptoms such as urinary complaints and pelvic pain, while rare cases involving pelvic nodes have been reported in the past [13]. Due to non-specific symptoms and clinical presentation, the differential diagnosis of endocervicosis is difficult: since it is commonly associated with a mass, it may be confused with a malignant tumor and therefore it must be clinically distinguished from an invasive carcinoma. Cystoscopy, which is still the gold standard investigation for the differential diagnosis of bladder cancer, invariably shows a mural lesion covered by intact epithelium. The definitive diagnosis relies on careful histopathological examination of the resected tissues. The histological differential diagnosis of endocervicosis includes several benign and malignant conditions such as cystitis glandularis, cystitis cystica nephrogenic adenoma and well-differentiated adeno-carcinoma of the bladder [3]. The final pathological diagnosis is based on architectural pattern and cytological features of the lesion.

According to Rodriguez *et al,* partial cystectomy represents the best therapeutic option for patients with endocervicosis of the bladder [12]. A successful laparoscopic excision of endocervicosis of the urinary bladder has also been reported [14]. Less invasive therapeutic approaches have also been suggested [13], while other authors, due to the benign condition of the lesion, proposed an even less invasive approach, rejecting the complete transurethral resection [15]. According to the author's point of view, symptomatic endocervicosis can be managed with complete transurethral resection in order to reduce the burden of symptoms.

Fewer than 25 cases of endocervicosis of the bladder have been reported and therefore the natural history of the disease is unknown. However, there is no documented case of recurrence in a 14-year follow-up interval [2]. According to the perspective of the authors, the rarity of the condition and the sparse information on long term follow up, dictates that the follow-up should be continued indefinitely.

Conclusion

Endocervicosis of the bladder wall is a rare lesion, and the diagnosis is difficult to make both clinically and pathologically. Important aids to distinguish this entity from bladder cancer are adequate biopsies with close correlation between the urologist and histopathologist. It is important that this lesion be recognized and not misdiagnosed as a malignant neoplasm. Despite the rarity of the disease, urologists should be aware of this condition for proper diagnostic and surgical management.

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