Case report

Detrusor instability is a rare presentation of pelvic subserosal fibroma, seven years following a total abdominal hysterectomy and bilateral salpingo-oophorectomy: Case report

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Abstract: Solitary Fibrous Tumours (SFT) are uncommon, slow growing, well-circumscribed tumours arising from the mesenchyma. These tumours were first described in the thoracic cavity, originating from the pleura but have since been reported in various sites including the prostate, bladder, periosteum, soft tissue, liver and kidney. This case report describes a 40 year old woman, presenting with symptoms of detrusor instability due to a midline solid, discrete pelvic mass, centred above the bladder, with displacement of the sigmoid colon evident on CT scan. Histology revealed a 'subserosal fibroma'. This article describes the current literature relating to pelvic solitary fibrous tumours.

Key words: Subserosal fibroma; Solitary fibrous tumours; Detrusor instability

INTRODUCTION

Solitary fibrous tumours (SFT) or commonly known as fibromas, were first documented at the turn of the 20th century, originating in the intrathoracic cavity, typically from the pleura. Fibromas are slow growing, well circumscribed, spindle cell neoplasms arising from the mesenchyma. Since the 1960s, there have been an increasing number of case reports describing fibromas in extrathoracic sites including the orbit, meninges, upper respiratory tract, thyroid, salivary gland and spinal cord. In the last 20 years, reports of fibromas have been documented in the peritoneum, retroperitoneum, urinary bladder, uterus, cervix, vulva, vagina, paravaginal space, ischiorectal fossa and fallopian tube[1-8].

The presentation of pelvic solitary fibrous tumours remains highly variable and related to anatomical relationships and size[4]. SFT are commonly asymptomatic, but compression on adjacent structures has been associated with bowel obstruction and bilateral ureteral obstruction[4], vaginal bleeding or dyspareunia[10]. There have been case reports of systemic features such as arthralgia and hypoglycaemia in cases of extrathoracic SFT[1,5,8]. This case report describes a unique presentation of a pelvic fibroma with detrusor instability following a total abdominal hysterectomy and bilateral salpingo-oophorectomy.

The search strategy used in Medline included the key words; solitary fibrous tumour, pelvis and subserosal and the subject headings; neoplasms fibrous tissue and fibromas. Hand searching methods were also employed. There were 14 english articles pertaining to solitary fibrous tumours in the pelvis.

CASE REPORT

A 40 year old woman was referred by her general practitioner (GP) because of stress urinary incontinence, urgency and urge incontinence. She was seen in the urogynaecology clinic and stated that these symptoms started few years ago but have worsened recently. She passes urine up to 8 times during the day and once during the night. Seven years ago she had total abdominal hysterectomy and bilateral salpingo-oophorectomy for pelvic endometriosis, confirmed by histological examination. It was stated in the operative notes that no residual disease was left. She had been on annual oestradiol implants since her procedure. Recent testing of midstream urine sample arranged by her local doctor did not show any urinary tract infection. She had two children via normal vaginal delivery. There was no other past medical or surgical history of significance.

On examination, she was generally well with abdominal palpation normal. Pelvic examination revealed a normal vulva and vagina with no genital prolapse. Bimanual pelvic examination revealed a large solid mass that was felt centred in her pelvis. The urodynamic assessment revealed no residual urine with normal uroflowmetry. Detrusor instability was demonstrated and associated with significant urgency but no urge incontinence. No urodynamic stress urinary incontinence was demonstrated. A pelvic ultrasound was arranged and a blood test to check for serum tumours markers (CA 125, CA 19.9 and CEA).

On her follow-up visit, all tumour markers were well within normal limits. The pelvic ultrasound showed a midline solid pelvic mass measuring 9.1 x 8.5 x 8.4 cm. There was no evidence of hydronephrosis in either kidneys and no ascites. Due to uncertainty of the origin of the mass, a CT scan of the pelvis and abdomen was arranged.

Portal venous and delayed phase multi-detector spiral CT scan showed a solid pelvic mass centred to the right of midline but occupying the greater part of the lesser pelvis. It measured 10.7 x 8.5 x10.2 cm. It was centred above the bladder but separate from it. The mass displaced the sigmoid colon superiorly and was adjacent to the vaginal vault. No definite organ of origin could be identified. There was very small volume of ascites but no intra-abdominal lymph node enlargement. The kidneys, liver, spleen, pancreas and adrenal glands were normal. The CT images of from this case study are shown in Figure 1.

Due to the past history of the total abdominal hysterectomy and bilateral salpingo-oophorectomy along with the uncertainty of the origin of the mass she was referred to the surgical team. The findings from the laparotomy by the surgical team revealed a large pelvic mass, which was adhered to the sigmoid colon. The procedure was complicated by the presence of vascular adhesions. The bowel was separated from the mass uneventfully and the mass was completely removed. The histology of the mass was reported as a subserosal fibroma with no malignant changes seen.

The patient made a complete recovery. No postoperative urodynamic assessment was performed due to complete resolution of her urinary symptoms following her surgery. At the 2 year follow up appointment, she remained symptom free.

DISCUSSION

This case report outlines the significance of considering solitary fibrous tumours as a differential diagnosis for a pelvic mass resulting in common urogynaecological presentations. The differential diagnosis of a pelvic fibroma most commonly includes leiomyoma or leiomyosarcoma, hemangiopericytoma and fibrosarcoma[3].

In 1997, Chan proposed diagnostic criteria for solitary fibrous tumours, with the essential diagnostic features

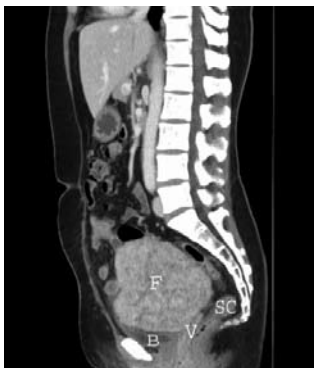

Fig. 1. – Is a sagittal section from the portal venous and delayed phase spiral computerised tomography of the patient in this case study. This figure illustrates clearly the subserosal solitary fibrous tumour in the paravaginal space, centered above and compressing the bladder along with displacing the sigmoid colon. The mass measures 10.7 cm anterioposteriorly x 8.5 cm wide x 10.2 cm craniocaudally (SC: sigmoid colon, B: bladder, V: vagina and F: fibroma).

including circumscription, hypercellular and hypocellular foci, short spindly with scanty and poorly defined cytoplasm, haphazard, storiform or fascicular arrangement of spindle cells, interwining of thin or thick collagen and CD 34 positive. Immunohistochemical markers such as CD 34 are used to further differentiate solitary fibrous tumours from myofibromas, neurofibromas, nodular fasciitis, dermatofibroma and dermatofibrosacroma protuberans[10]. The CD 34 antigen is a transmembrane glycoprotein on the cell surface that is widely used as a vascular marker[12]. Positive markers for solitary fibrous tumours include bcl-2, CD 99 and CD 34.

The majority of extrathoracic solitary fibrous tumours follow a benign course. Chan and colleagues found that malignancy was documented in13% of extrathoracic SFT, compared with the 23% of intrathoracic tumours. A review article examining the histological features and outcome of 7 cases of retroperitoneal solitary fibrous tumours found there was local recurrence in one case, associated with incomplete resection[3]. Chan believes this observed difference is due to the more recent recognition and reporting of the extrathoracic solitary fibrous tumours, rather than a lower malignant potential. Therefore, histopathology can not always predict the clinical behaviour, and follow up is essential[12].

The importance of imaging of palpable pelvic masses is essential in the diagnosis and planning for surgical management (see Figure 1 and 2). MR imaging of the pelvic region may be useful in further distinguishing the tissue of origin and has an important role in staging of pelvic tumours, although imaging modalities have not been specifically defined for solitary fibrous tumours[13]. Colour or Power Doppler ultrasound and MRI have been shown to further differentiate subserosal myomas from extrauterine tumours[14]. Interestingly, one case report describes central malignant degeneration of an extrathoracic SFT in the pelvis of a 61 year old man, which was detected on CT and MRI[15].

In this case study, the significance of the previous total abdominal hysterectomy and bilateral salpingo-oophorectomy 7 years prior to this presentation is most probably unrelated to the subserosal fibroma but contributed to a more complicated surgical resection. There has been only one case report in the literature that described a benign solitary uterine leiomyoma on the pelvic peritoneum following a hysterectomy for subserosal fibroids 5 years previously[16].

The ability to resect the extrathoracic solitary fibrous tumours is the major determinant of prognosis and ongoing surveillance is essential[12]. While the recognition of extrathoracic solitary fibrous tumours has evolved in the last 20 years, the epidemiology, pathophysiology and long term prognosis of this mesenchymal tumour remains uncertain.

Most cases of detrusor instability are idiopathic in nature. When a pelvic tumour compresses on the bladder and causes urinary symptoms, urodynamic assessment is more likely to reveal stress urinary incontinence rather than detrusor instability. Pelvic tumours can irritate the bladder and result in detrusor instability.

No postoperative urodynamic assessment was performed in this patient as she continued to be asymptomatic until she was last seen 2 years after her surgery.

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