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CASE REPORT

Large paravaginal solitary fibrous tumor with secondary schistosoma hematobium infestation

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Schistosomiasis of the female pelvic organs has been frequently reported in endemic areas. Cervix, vaginal wall and vulva are more frequently affected than fallopian tubes, the ovaries and uterus. The disease manifests itself as ulcers, polyps, warts, granulomatous lesions and small pelvic nodules (1, 2).

Paravaginal tumors are extremely rare with most of these tumors being of mesenchymal, in particular smooth muscle, neural and gastrointestinal stromal tumors, and including a wide variety of lesions like lipomas, fibrosarcomas, neurofibromas, and neurilemomas, dermoid cysts, leiomyosarcomas and malignant peripheral nerve sheath tumors (3).

To the best of our knowledge, we report the first case in the medical literature of a large Paravaginal solitary fibrous tumor that was secondarily infested by numerous schistosoma hematobium eggs, which have also induced a florid granulomatous reaction.

Case report

A 37-year old woman, para 7+0, was referred to our gynecological clinic in April 2000. Her local gynecologist identified a large pelvic mass for which she had had laparotomy for a presumed ovarian cyst, during the second trimester of her last pregnancy. At laparotomy, a large cystic tumor was found arising from the pararectal and paravaginal spaces with no relationship to the ovaries. Incisional biopsy was performed, and 150 ml of straw colored fluid was aspirated transvaginally from the tumor cavity; cytological and histological examination demonstrated only fibrous tissues, and inflammatory cells with no evidence of malignant cells. Her last pregnancy ended by cesarean section due to obstruction of the birth canal by the tumor.

The physical examination at the time of presentation to our hospital was normal, apart from the pelvic examination, which revealed a large, moderately firm mass almost obstructing the vaginal cavity. The cervix could not be visualized, the mass was not adherent to the pelvic wall, and the rectal and vaginal walls slipped freely over it. Transvaginal ultrasound showed a pelvic

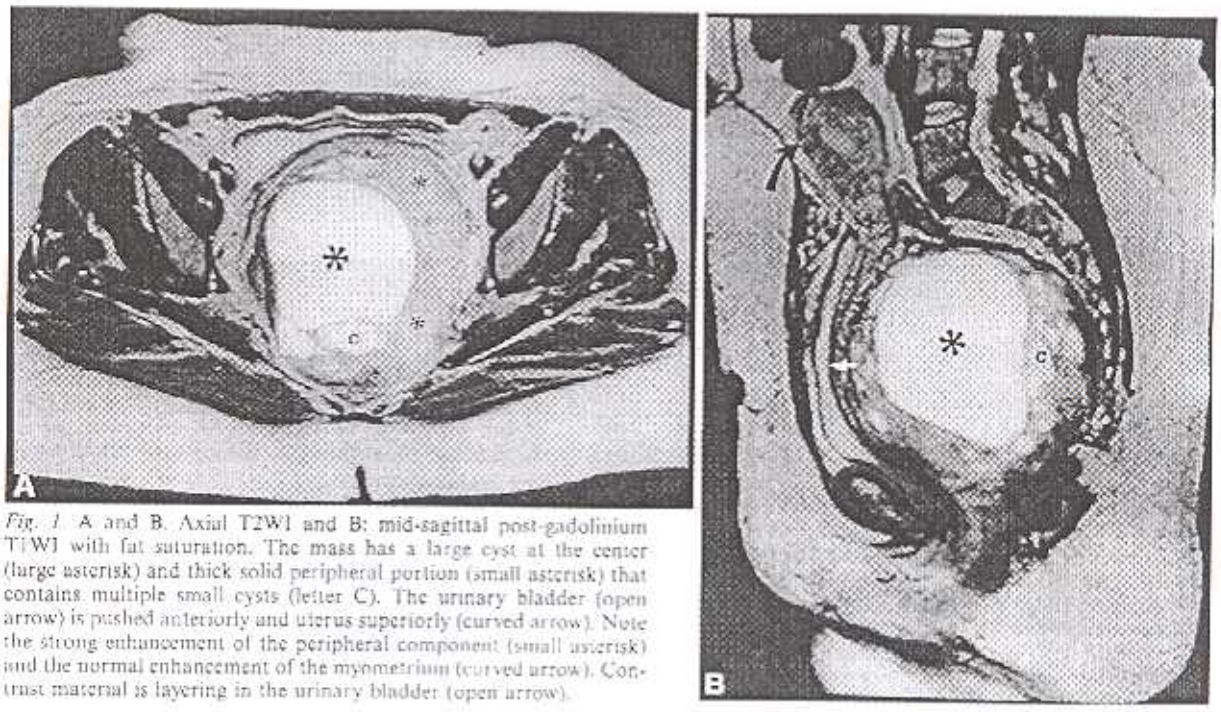

Fig. 1. A and B. Axial T2WI and B: mid-sagittal post-gadolinium T1WI with fat saturation. The mass has a large cyst at the center (large asterisk) and thick solid peripheral portion (small asterisk) that contains multiple small cysts (letter C). The urinary bladder (open arrow) is pushed anteriorly and uterus superiorly (curved arrow). Note the strong enhancement of the peripheral component (small asterisk) and the normal enhancement of the myometrium (curved arrow). Contrast material is layering in the urinary bladder (open arrow).

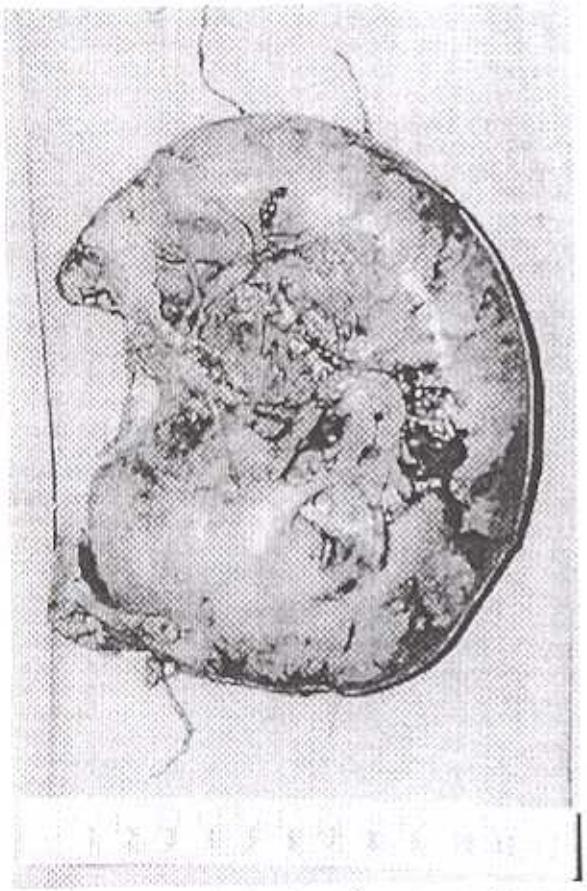

Fig. 2. Large paravaginal solitary fibrous tumor showing a smooth pale and partly hemorrhagic outer surface.

mass of mixed echogenicity, with a cystic center pushing the uterus upwards and anteriorly. Magnetic resonance imaging (MRI) revealed a large pelvic mass, measuring 13.0×12.0×11.1 cm, composed of a cystic center and solid periphery which contained multiple smaller cysts (Fig. 1A and 1B). The uterus and urinary bladder were pushed superiorly above the pelvic brim; vagina was stretched and displaced anteriorly while the rectum was displaced posteriorly. There was no evidence of invasion of the surrounding structures and no pelvic or intraabdominal lymphadenopathy was noted. Chest x-ray was normal. The working differential diagnosis included leiomyoma and fibroma with necrotic center. Because of the large size of the tumor, a combined vaginal and abdominal surgical approach was selected. An exploratory laparatomy was performed; uterus, tubes, and ovaries were normal, after aspiration of 350 ml of straw colored fluid; the pararectal and paravaginal space was entered through the Pouch of Douglas, and the upper pole of the tumor was shelled out of its bed by blunt dissection. Through a vaginal incision, the lower pole of the tumor was bluntly dissected from the surrounding tissues and the mass was then delivered transvaginally. Postoperative repeated urine and stool microscopical examination showed no evidence of schistosomiasis, but immunological tests for schistosoma were positive. The patient received a single dose of praziquantel (40 mg/kg) orally. The postoperative recovery was uneventful.

Pathological findings

On gross examination, the excised mass measured 12.0×10.5×5.0 cm (Fig. 2). The cut surface showed a large hemorrhagic cavity near the center with surrounding solid and pale tissue. The histological sections showed an inflammatory fibrovascular mass containing numerous schistosoma hematobium eggs with terminal spines surrounded by many epithelioid and giant cell granulomas with a large number of eosinophils (Fig. 3A and 3B). The described granulomas were set within a benign fibrous tumor consisting of numerous bland spindle cells, which are arranged haphazardly in a densely collagenous matrix containing many blood vessels and a central necrotic area; most probably representing ischemic necrosis with hematoma formation due to previous aspiration. There was no evidence of increased mitotic activity or malignancy. Immunohistochemically, the tumor cells were strongly positive for CD34 and vimentin but negative for S100 protein, desmin, and smooth muscle actin; thus the mass was diagnosed as a solitary fibrous tumor.

Discussion

Paravaginal tumors are rare neoplasms of the female genital tract. The differential diagnosis usually includes a wide variety of retroperitoneal and intraperitoneal tumors. Most of these tumors are malignant with sarcomas being the commonest

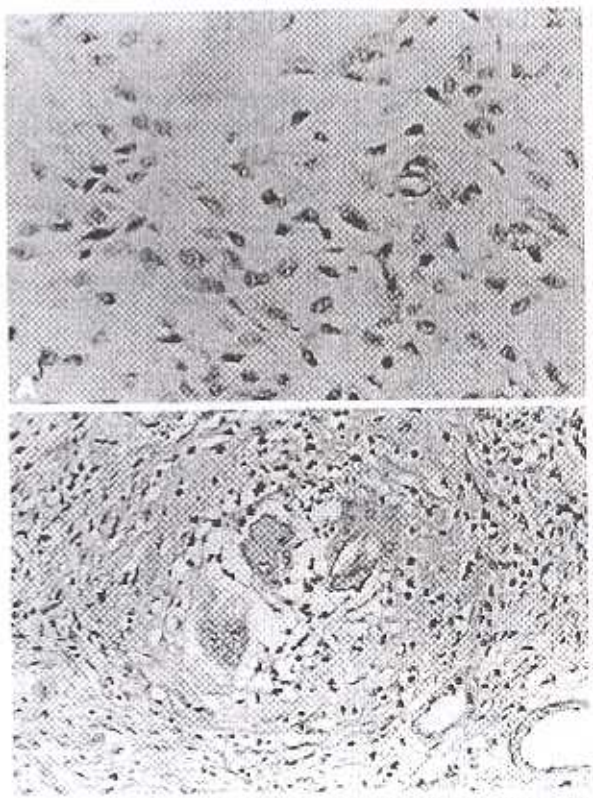

Fig. 3. A. Histological section of paravaginal solitary fibrous tumor showing many cells with oval and spindle shaped nuclei in a loose and fibrovascular connective tissue background. Hematoxylin and eosin stain ×400. B. A giant cell granuloma containing a 'shrunken' engulfed schistosoma egg with surrounding fibrosis and inflammation. Hematoxylin and eosin stain ×200.

retroperitoneal pelvic tumors (3). Solitary fibrous tumors are first described as pleural lesion, but increasing number of extrathoracic sites including kidneys, neck, groin, buttocks, retroperitoneum and orbital lesions have been reported (4–7). Extrapleural lesions, irrespective of site, almost invariably arise in adults of either sex who present with a nondescript slowly enlarging mass and the clinical course of most of solitary fibrous tumors seem to be benign (6). Although their clinical outcome is difficult to predict, solitary fibrous tumors with atypia, necrosis, hypercellularity and greater than 4 mitosis/HPFs are associated with, but not predictive of, aggressive behavior (7).

Schistosoma hematobium adult worms are well known to lay their eggs in mesenteric veins (1, 2). Vascularity of solitary fibrous tumors is typically rich. These characteristics of both the parasite and tumor may explain the combination of schistosoma hematobium and paravaginal solitary fibrous tumors. The large size of this tumor, the result of immunohisto chemical tests, and the lack of documented evidence in the medical literature of similar schistosoma induced complications indicate that the most probable diagnosis is secondary schistosoma infestation of pre-existing solitary fibrous tumor. Our case also illustrates the importance of preoperative assessment of retroperitoneal pelvic tumors, as inadequate preoperative diagnosis led to a mistaken diagnosis of ovarian cyst with inappropriate intervention during pregnancy. Schistosoma hematobium infestation of pelvic tumors is an uncommon phenomenon but may occur in patients living in endemic areas. Praziquantel is a safe and highly effective agent against all schistosoma species and is available orally. The behavior of Paravaginal solitary fibrous tumor is unpredictable, therefore complete surgical excision with clear margin, and long-term follow-up for patient is advisable (6, 7).

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