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Case Report

Sacrococcygeal Neurofibroma: Rare Cause for Chronic Pelvic Pain

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ABSTRACT Pelvic pain is a common gynecologic complaint. Retroperitoneal pelvic tumors are rarely a cause of pelvic pain. Neurofibroma is an uncommon pelvic retroperitoneal tumor, and only 17 cases are reported to date. A 38-year-old woman with chronic pelvic pain had a soft fixed mass that was the size of an orange in the right posterolateral fornix, with a normal uterus on pelvic examination, and a mass of 6.3 × 5.2 cm with mixed echotexture on the right side separate from both ovaries on transvaginal ultrasonography. A provisional diagnosis of retroperitoneal mass probably a retroperitoneal teratoma was made. Laparoscopy was performed; an ill-defined retroperitoneal soft tissue mass of about 6 cm was seen on the right pararectal and presacral area, displacing the rectum toward the left side. The mass was soft and jellylike without a cyst wall. Histopathologic study and immunohistochemistry results were consistent with neurofibroma of the sacrococcygeal regions. To our knowledge this is the third case of sacrococcygeal neurofibroma treated by complete laparoscopic excision. Gynecologists should keep sacrococcygeal neurofibroma as a differential diagnosis of pelvic pain with atypical location of a pelvic mass. A high index of suspicion and an appropriate imaging technique are needed for accurate diagnosis. Laparoscopy seems to be a safe and effective method of managing retroperitoneal presacral neurofibromas. Journal of Minimally Invasive Gynecology (2012) 19, 517–520 © 2012 AAGL. All rights reserved.

Keywords: Sacrococcygeal neurofibroma; Chronic pelvic pain; Laparoscopic surgery

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Pelvic pain is a common gynecologic complaint. Diagnosing the cause of pelvic pain is straightforward in most cases. Rarely retroperitoneal pelvic tumors can be a cause for pelvic pain. The most common pelvic retroperitoneal tumors are lymphosarcomas [1]. Neurofibroma is an uncommon pelvic retroperitoneal tumor, and only 17 cases are reported to date [2]. Diagnosis of retroperitoneal tumors is difficult unless there is a high index of suspicion. We report a case of retroperitoneal presacral neurofibroma in a patient who presented with pelvic pain that was excised laparoscopically. To our knowledge this is the third case of sacrococcygeal neurofibroma treated by complete laparoscopic excision [3,4].

Case Report

A 38-year-old woman, para 2, consulted our hospital for pain of 6 months duration in the lower abdomen. Her menses were regular and painless. Ultrasonography of the pelvis performed elsewhere was suggestive of a complex right ovarian cyst.

Pelvic examination showed a soft fixed mass that was the size of an orange in the right posterolateral fornix with a normal uterus. Transvaginal ultrasonography showed a mass of 6.3 × 5.2 cm with mixed echotexture on the right side separate from both ovaries. There was no abnormal vascularity on color-flow Doppler study. A provisional diagnosis of retroperitoneal mass, probably a retroperitoneal teratoma, was made.

Laparoscopy was performed with a 4-puncture technique. The uterus was normal in size, and both tubes and ovaries were normal. An ill-defined retroperitoneal soft tissue

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Fig. 1

Sacrococcygeal neurofibroma and its relation to pelvic organs.

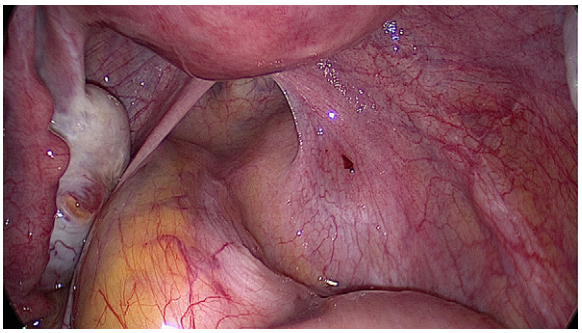
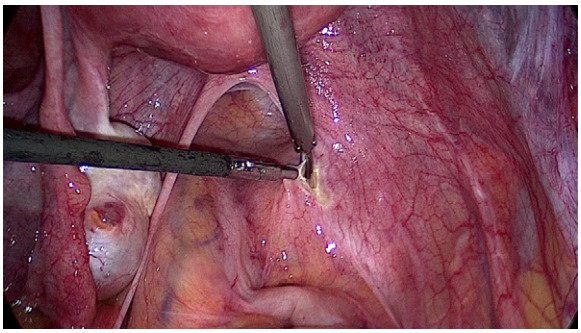


Fig. 3

Opening of peritoneum overlying neurofibroma with Harmonic Ace.



mass of about 6 cm was seen on the right pararectal and presacral area, displacing the rectum toward the left side. The mass was not adherent to any pelvic organs. No ligamentous attachment or vascular pedicle was seen (Figs. 1 and 2). An oblique incision was made on the peritoneum overlying the mass with Harmonic Ace (Ethicon Endo-surgery, Johnson and Johnson Medical, New Brunswick, NJ) after identification of the right ureter (Fig. 3). The rectum was identified and pushed away by a rectal probe. The mass was enucleated with difficulty by blunt and sharp dissection, because it was soft and difficult to hold with a grasper. The mass was soft and jellylike without a cyst wall (Fig. 4). There was no neural attachment or vascular pedicle to the mass. The excised site was examined for completion and hemostasis (Fig. 5). Because the patient requested sterilization, Fallope rings were applied on both tubes. A specimen was removed through a 10-mm primary port and visualized with a 5-mm telescope introduced through one of the accessory ports. A drain was kept at the surgical site for 8 hours. Total duration of the surgery was 60 minutes. The patient's recovery was uneventful, and she was discharged after 24 hours. At 6 months follow-up, the patient was pain free, without any neurologic deficit.

Histopathologic study showed tissue pieces of a nonencapsulated neoplasm composed of loosely spaced spindle cells and wavy collagenous strands. The spindle cells displayed wavy nuclei with tapered ends and scant cytoplasm. No significant cytologic atypia/mitoses were seen (Fig. 6). The results of immunohistochemistry were as follow: the spindle cells were S100, Vimentin was positive, neuron-specific enolase was focally positive, and green florescent protein and smooth muscle actin were negative. The MIB-I proliferation index was very low. Immunohistochemistry and histopathologic study results were consistent with neurofibroma of sacrococcygeal regions (Fig. 7).

Discussion

Chronic pelvic pain has been defined as pain of greater than 6 months' duration, localized to the anatomic pelvis, and severe enough to cause functional disability or necessitating medical care [5]. Chronic pelvic pain is estimated to account for 10% of all referrals to gynecologists. It is the indication for 12% of all hysterectomies and more than 40% of gynecologic diagnostic laparoscopies [5]. Approximately 60% to 80% of patients undergoing laparoscopy for chronic pelvic pain have no intraperitoneal disease, nor do they have tissue distortion that correlates with the pain [6]. Often the cause of chronic pelvic pain is not clear, because there are many disorders of the reproductive tract, gastrointestinal system, urologic organs, musculoskeletal system, and

Fig. 2

Sacrococcygeal neurofibroma and its relation to pelvic organs.

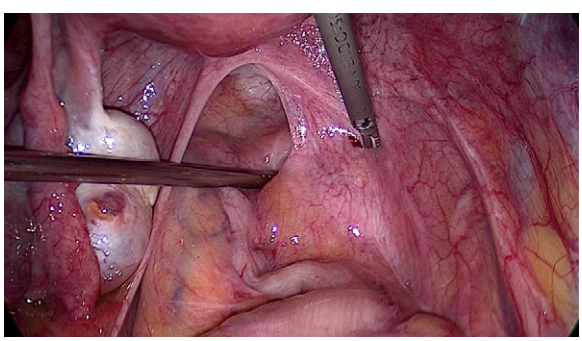
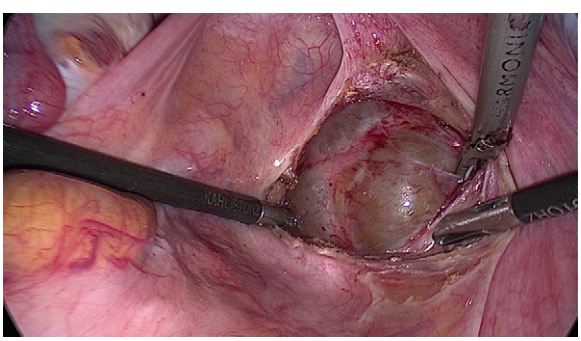


Fig. 4

Enucleation of neurofibroma.



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