

A case report of primary multiple obturator nerve schwannomas

Journal of International Medical Research 48(9) 1–4 © The Author(s) 2020 Article reuse guidelines: sagepub.com/journals-permissions DOI: 10.1177/0300060520959490 journals.sagepub.com/home/imr



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Abstract

Primary multiple obturator nerve schwannomas originate from Schwann cells and are extremely rare. Patients with schwannomas are asymptomatic and a retroperitoneal schwannoma is often misdiagnosed as an adnexal mass. In the present study, we describe a 58-year-old woman in whom a right adnexal mass accompanied by endometrial polyp was found incidentally through transvaginal ultrasound. The mass was diagnosed as multiple obturator nerve schwannomas after laparoscopy. Immunohistochemical assay confirmed the schwannomas to be positive for SOX10. To our knowledge, this is the first report to demonstrate a case of multiple schwannomas originating from the obturator nerve and treated by laparoscopic resection.

Keywords

Multiple schwannomas, obturator nerve, adnexal mass, laparoscopy, immunohistochemistry, SOX10

Date received: 13 March 2020; accepted: 27 August 2020

Introduction

Schwannoma is a well-encapsulated tumor composed of peripheral Schwann cells. As a type of benign nerve sheath tumor, schwannoma mostly occurs in the cranial nerves or the nerves of the upper extremities; only about 3% of schwannomas exist in the retroperitoneum.¹ The majority of retroperitoneal schwannomas are asymptomatic; they are usually found incidentally and

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misdiagnosed as gynecological masses or as urological disease.² Tumor resection and histopathology examination are essential for proper diagnosis. This is the first report describing successful laparoscopic resection of multiple obturator nerve schwannomas without nerve complications.

Case presentation

A 58-year-old woman was hospitalized because of a right adnexal mass accompanied by endometrial polyp on July 12, 2019. She had no symptoms of discomfort, and laboratory data were all unremarkable after admission. Transvaginal ultrasound demonstrated a hypoechoic mass of $4.06 \times$ 2.09 cm in the right adnexal area, uneven echo, and dotted blood flow signals (Figure 1a). Contrast-enhanced computed tomography (CT) confirmed a nonenhancing cystic mass of $30 \times 28 \text{ mm}$ (Figure 1b). The patient consented to and underwent a laparoscopic bilateral tubal oophorectomy combined with hysteroscopy.

The patient was operated on in the lithotomy position after introduction of general anesthesia. An endometrial polyp of 1.5×0.8 cm was observed and removed entirely under hysteroscopy. Laparoscopy revealed that the right ovary was cystic

and enlarged (approximately $4.0 \times 3.0 \times$ 1.0 cm), and the left ovary and uterus were atrophic. After a conventional bilateral tubal oophorectomy, a bulge was seen on the right wall of the pelvic cavity (Figure 2). The right posterior peritoneum was opened using a harmonic scalpel; the ipsilateral ureter, iliac arteries, and corresponding veins were identified and dissociated; and gelatinous masses grew around the obturator nerve like a string of beads. Of these masses, the largest was 4.0×3.0 cm and the smallest was 0.5×0.5 cm. Frozen analysis revealed the masses were benign schwannomas, and they were removed carefully by means of the harmonic scalpel. The integrity of obturator nerve was preserved and thorough hemostasis ensured during the operation. The patient tolerated the surgery well and was discharged on postoperative day five.

Final histopathology of the masses showed solitary, well-circumscribed, encapsulated neoplasms; small vessels and glands were observed. The typical schwannoma histological structures of Antoni A and B patterns were observed. Areas of higher and more compact cellularity corresponded to the Antoni A patterns, whereas areas of looser cellularity corresponded to Antoni B patterns. Immunohistochemical examination revealed strong positive staining for

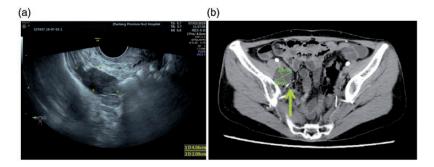


Figure 1. Imaging examination of the right adnexal mass. (a) A hypoechoic mass of 4.06×2.09 cm observed in right adnexal area. (b) The pelvic mass is identified as a non-enhancing mass of 30×28 mm adjacent to the right pelvic wall.

SOX10 protein (Figure 3). These results were consistent with the histological diagnosis of benign schwannomas.

Discussion

A literature search of the PubMed database using the medical subject headings (MeSH) terms "pelvic schwannoma" and "obturator nerve" indicated that this case is the first reported case of multiple obturator nerve schwannomas. Schwannomas are exceedingly rare and slow-growing tumors that originate from neuronal sheath cells.³ Some patients with obturator nerve schwannoma present with mild abdominal pain or distension, although most patients are asymptomatic. Ultrasound examination is usually the primary choice for evaluation because it is inexpensive and excellent at detecting cystic lesions. However, because schwannoma is rare, the accuracy of ultrasound examination is limited. Contrastenhanced CT plays a vital role in localizing and characterizing tumors, assessing local invasion and metastases, and determining the treatment response of tumors.⁴ The final diagnosis depends on histopathology.

In the published cases, the benign schwannomas were all found as a single occurrence, and complete resection was recommended.⁵ However, multiple masses were identified in our case and it was essential to distinguish whether they were schwannomas or neurofibromas. The treatment modality for schwannomas and

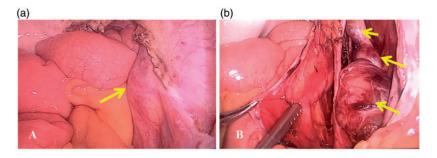


Figure 2. Laparoscopic resection of the obturator nerve schwannomas. (a) The arrow indicates a bulge behind the right posterior peritoneum. (b) The arrows indicate multiple schwannomas, which are well-encapsulated and located in the right obturator nerve.

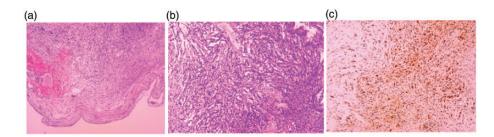


Figure 3. Histopathology findings of the obturator nerve schwannomas. (a) The masses are encapsulated by an integrated envelope, and small blood vessels can be observed (hematoxylin and eosin, $50 \times$ original magnification). (b) The areas of higher, compact cellularity are Antoni A areas, and the areas of looser cellularity are Antoni B (hematoxylin and eosin, $100 \times$ original magnification). (c) Strong immunoreactivity of SOX10 protein confirmed the masses to be schwannomas ($100 \times$ original magnification).

neurofibromas is completely different: conservative observation instead of surgery is recommended for neurofibromas. By immunohistochemical examination. schwannomas show diffuse reactivity of SOX10 or S100 protein, where neurofibromas show only sporadic expression of SOX10 or S100.⁶ SOX10 has recently been shown to have better diagnostic performance than other classical markers.⁷ Scotto et al. reported a patient whose obturator nerve was sectioned accidentally and then anastomosed with Prolene 6/0 suture; the patient eventually recovered without sensory or motor sequelae.⁸ Therefore, tumor resection is recommended despite the risk of nerve damage because the prognosis following resection is good.

Conclusion

We reported a case of benign multiple obturator nerve schwannomas. Preoperative diagnosis can be difficult because the disease lacks distinctive clinical symptoms and specific imaging findings. Intraoperative analysis of a frozen section helps to accurately identify tumor characteristics and confirm the diagnosis, and surgical resection is recommended for the treatment of schwannoma.

Ethical approval

This study and publication were approved by the ethics committee of the 2nd Affiliated Hospital, Zhejiang University School of Medicine (Hangzhou, People's Republic of China). The patient provided written informed consent for publication of this paper.

Declaration of conflicting interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This work was supported by grant from the National Natural Science Foundation of China (Grant No. 81472422).

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