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Presacral Schwannoma Causing Infertility in a Young Woman: Report of a Case

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Introduction: Schwannoma is one of the tumors that may occur in the sacral or presacral area. It is often asymptomatic and found incidentally by obstetricians or other clinicians. In this study, we report a case of presacral schwannoma as a rare cause of infertility.
Case Presentation: A 33 year-old woman referred to a gynecology clinic with complain of two years infertility and one abortion. All examinations and clinical evaluations were normal except one homogenous mass on transvaginal ultrasound. After resection via a transabdominal approach her infertility resolved after thirteen months. Histopathologic examination revealed benign schwannoma.
Conclusion: Presacral schwannoma should be considered as a rare cause of infertility which could be resolved successfully after operation.

Keywords: Neurilemmoma; Infertility; Iran

1. Introduction

Occurance of presacral (retrorectal) tumors in adults is rare. Although the majority of them are benign, they may cause a number of problems in diagnosis and management (1). The incidence rate of presacral tumors in the general population is not known because the majority of reports on these lesions are from tertiary referral centers (2). The only large serious occurrence of presacral tumor-not from a referral center-was published by Hobson in 2005 that found an average incidence of two presacral tumors per year in the metropolitan Portland area (3). Among other pathologic types (neurofibromas, neurofibrosarcomas, ependymomas, chordomas), benign schwannomas (neurilemmomas) are classified as neurogenic tumors that occurred in the sacral or presacral space (1)(4). Schwannomas are generally benign, slow-growing and painless tumors arising from Schwann cells or peripheral nerve sheaths (5-7). Although the tumor arises from the peripheral nerve, it rarely elicits clinically detectable neurological deficits. Schwannomas found in the thoracic cavity, retroperitoneum, or pelvis are usually quite large with involvement of adjacent visceral structures (6). The low incidence of this tumor and the lack of specific signs and symptoms make preoperative diagnosis very difficult (1).

2. Case Presentation

A 33 year-old woman referred to a gynecology clinic following two years infertility. All obstetric examinations and laboratory tests were normal except a well circumscribed, homogenous mass adjacent to but separate from the right ovary that was found via transvaginal ultrasound. She was complained of low back pain but without any abdominal or pelvic pain, urinary complaints, or lower extremity numbness or weakness. During physical examination, her abdomen was soft and non-tender with no palpable masses. She mentioned a history of abortion at the first trimester.

Subsequent CT-scan with triple contrast revealed a well-defined oval shaped 56 × 48 mm solid mass in the right sacral area that showed enhancement at post contrast images. This was representing lymphadenopathy or neurogenic tumors (Figure 1).

Therefore pelvic MRI was recommended for further evaluation. Pelvic MRI showed a large unilobular mass measuring 68 × 47 × 42 mm at the anterior aspect of the right sacral ala that displaced the iliac vessels laterally representing benign neurologic tumors such as schwannoma (Figure 2).

Although appearance of the mass on imaging studies was consistent with a benign schwannoma, resection was suggested and the patient was convinced of her infertility as well as continued enlargement of the tumor over time. We decided to perform surgery using a transabdominal approach. The patient was taken to the operating room and a large pelvic mass was excised. At the time of surgery, the mass seemed to originate from the

Implication for health policy/practice/research/medical education:
This is a case study about unusual presentation of presacral tumors for improving knowledge of physician in better diagnose.

nerve root of the L5-S1 lumbar spinal space and it was easily dissected and removed from the sacrum posteriorly, with no nerve root involvement grossly. The tumor was well encapsulated without evidence of invasion to surrounding structures. Histopathologic examination of the resected specimen revealed interlacing bundles of elongated cells with spindle-shaped nuclei, diagnostic of a benign schwannoma. The patient was discharged to home on the fifth postoperative day without any complications. During our follow-up, no long term complications occurred. Thirteen months later, she experienced a normal pregnancy.

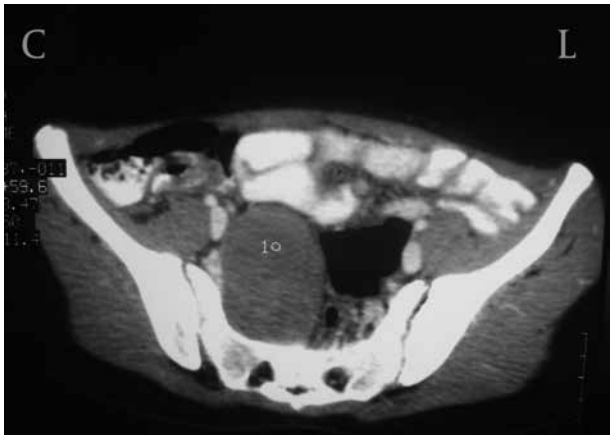

Figure 1. CT Scan Showing a 56 × 48 mm Mass in the Right Sacral Region

3. Discussion

The most common tumor of the peripheral nerves, particularly the cranial nerves, is the neurinoma, which is also called schwannoma because of its origin in the Schwann cells and presumably arising from a peripheral nerve in or adjacent to an anterior sacral foramen (7). Therefore, they are able to grow outside of the confines of the bone with limited degree of secondary bone involvement (6). Commencing from the peripheral epineurium, the tumor grows quite slowly, whereas localization and occurrence can be manifold (7). Principally, the tumor is found in the flexor group of the lower and upper extremities, in the head and neck region, or the trunk. In a literature, 36 cases involving localization of this tumor in the region of the retroperitoneum have been described. In these cases the schwannomas mimicked many different kinds of symptoms (8-11). The presacral retroperitoneum is a potential space. These are the possible reasons why these slowly growing tumors can develop to such a large size before symptoms occur clinically (6). They occur without gender predominance among the 20 to 50 year age group and are associated with von Recklinghausen's disease in 18% of cases. Malignant change is exceedingly rare; however, malignant tumors have been reported, although they are usually associated with von Recklinghausen's disease (10). The low incidence of this tumor and lack of specific sign and symptoms make preoperative diagnosis very difficult. They are often asymptomatic and found incidentally by obstetricians or other clinicians, as was the case of our patient (8, 12, 13). Radiological examinations such as ultrasonography, CT scan, or angiography usually cannot discriminate retroperitoneal schwannomas from other retroperitoneal tumors (6).

Treatment of schwannomas mainly consists of complete surgical excision. The prognosis is excellent (5). Extraperitoneal approach is reported to be effective in reduction of the chance of interfering with intestinal and gynecologic functions (13). We reported a rare case of schwannoma located in the presacral area; in which the clinical manifestation of infertility was more intersting. The surgical outcome was fair at 1 year postoperative follow-up and her infertility problem was resolved successfully.

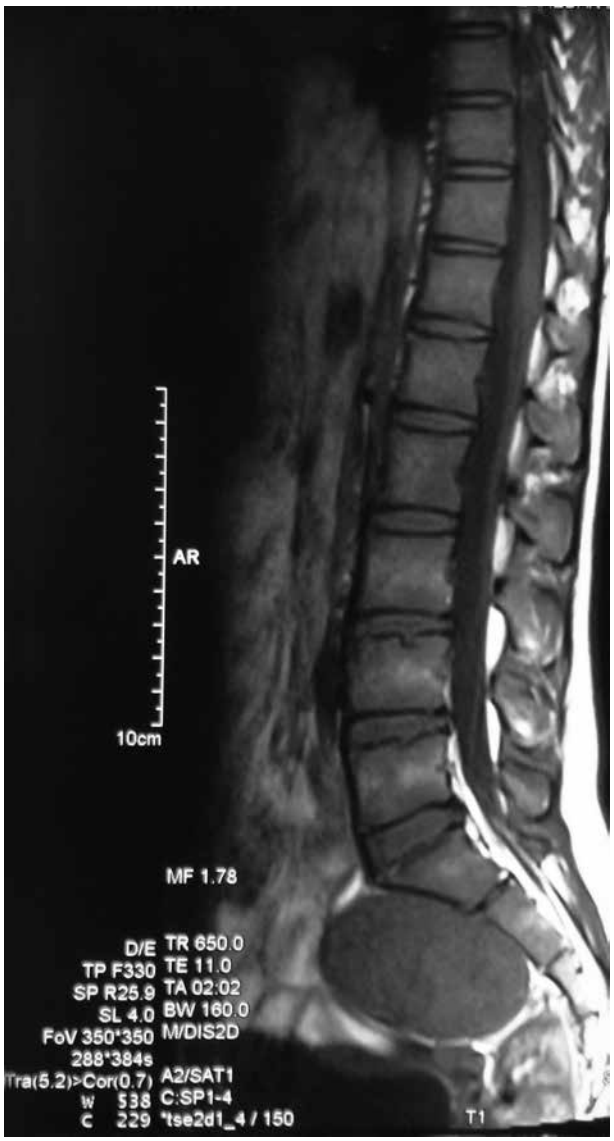

Figure 2. Sacral MRI Showing a Well-Defined Encapsulated Mass, in the Anterior Site of Right Sacral Area

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Authors' Contributions

Dr. Zamiri was the attending physician of this patient and Dr. Sarvestani collect the data, follow the patient and wrote this article.

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We do not have any financial disclosure and both authors were involved in management of this patient.

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