Perineural Spread Along Spinal and Obturator Nerves in Primary Vaginal Carcinoma: A Case Report

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Key words
- Imaging
- Lumbosacral plexopathy
- Perineural spread
- Primary vaginal cancer

Abbreviations and Acronyms
CT: Computed tomography
MRI: Magnetic resonance imaging

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Citation: World Neurosurg. (2018) 115:85-88. https://doi.org/10.1016/j.wneu.2018.04.030

Journal homepage: www.WORLDNEUROSURGERY.org

Available online: www.sciencedirect.com

BACKGROUND: **Perineural spread is not an uncommon feature in carcinoma of the head and neck. On the contrary, perineural spread in pelvic malignancies has been rarely reported. This is the first report on perineural spread of the obturator nerve and the lumbosacral plexus from primary vaginal cancer.**

CASE DESCRIPTION: **A 50-year-old woman diagnosed with infectious spondylitis at an outside hospital was referred to our institution. She presented with persistent lower back pain and right anterior thigh pain. Magnetic resonance imaging and subsequent ^{18}F-fluorodeoxyglucose-positron emission tomography/computed tomography revealed primary vaginal cancer with metastatic lymphadenopathy and perineural spread of the lumbosacral plexus, including L3, L4 nerve roots and branches, and obturator nerve along with soft tissue masses in the right psoas and proximal adductor muscles.**

CONCLUSIONS: **Clinical diagnosis of perineural spread in pelvic malignancies is difficult to establish, as it has nonspecific symptoms as an initial manifestation. Clinicians should recognize the possibility of perineural spread of malignancies if radiologic findings, such as thickening of lumbosacral nerves and perineural mass, are observed on magnetic resonance imaging. Further evaluation is necessary to detect possible pelvic malignancy and differentiate from other diseases.**

INTRODUCTION

Perineural spread in carcinoma of the head and neck is not unusual.[1,2] On the other hand, perineural spread of tumors in pelvic organs has been rarely reported. It has been described as a cause of lumbosacral plexopathy in cervical,[3] prostate,[4] bladder,[5] and rectal cancer.[6] To our knowledge there are no previous reports in the literature about the perineural spread from vaginal squamous cell carcinoma.

We report an unusual case of perineural spread of vaginal squamous cell carcinoma along the lumbar plexus and obturator nerve with extension into the psoas and adductor muscle compartments, initially mistaken for infectious spondylitis. We also describe its imaging findings on magnetic resonance imaging (MRI) and differential diagnosis.

CASE REPORT

A 50-year-old woman was admitted to our hospital with a 6-month history of persistent pain of the lower back and right anterior thigh. The patient initially visited an outside hospital because of right thigh and back pain 4 months before admission to our hospital. The patient had a history of spinal fusion (L4-5 level) and surgical incision and drainage due to surgical site infection 9 months earlier. Outside lumbar spine radiographs showed no abnormal finding. Nonenhanced lumbosacral spine computed tomography (CT) image showed edematous change of right psoas muscles, with perimuscular fat infiltration. Considering the clinical symptoms and past medical history of the patient, paravertebral infectious myositis with intramuscular abscesses was suspected. The patient underwent a sonography-guided aspiration in the right psoas muscle. Despite receiving the antibiotic treatment for 2 months, the patient complained of persistent pain and was referred to our institution.

On admission to our hospital, physical examination showed tenderness in her right anterior thigh, and decreased right knee jerk reflex, which is mediated by the L3 and L4 nerves. Complete blood count was within normal limit. The laboratory tests showed increased erythrocyte sedimentation rate and C-reactive protein.

The patient underwent surgical removal of implants to exclude infectious spondylitis. There was no evidence of bacterial infection in the culture of the surgical specimen. Lumbar spine MRI, taken just before surgery, demonstrated diffuse tubular thickening of right L3 and L4 nerves and perineural soft tissue masses encasing these abnormal nerves at right psoas muscles (Figure 1A and B). Additional pelvis MRIs were performed. MRIsdemonstrated diffuse thickening of the obturator nerve with perineural soft tissue masses throughout its course, which extends to L3 and L4 nerves (Figure 1C). Axial images at a more caudal level showed ambiguous anatomic boundary of the right-sided wall of the vagina, and continuity with a mass in

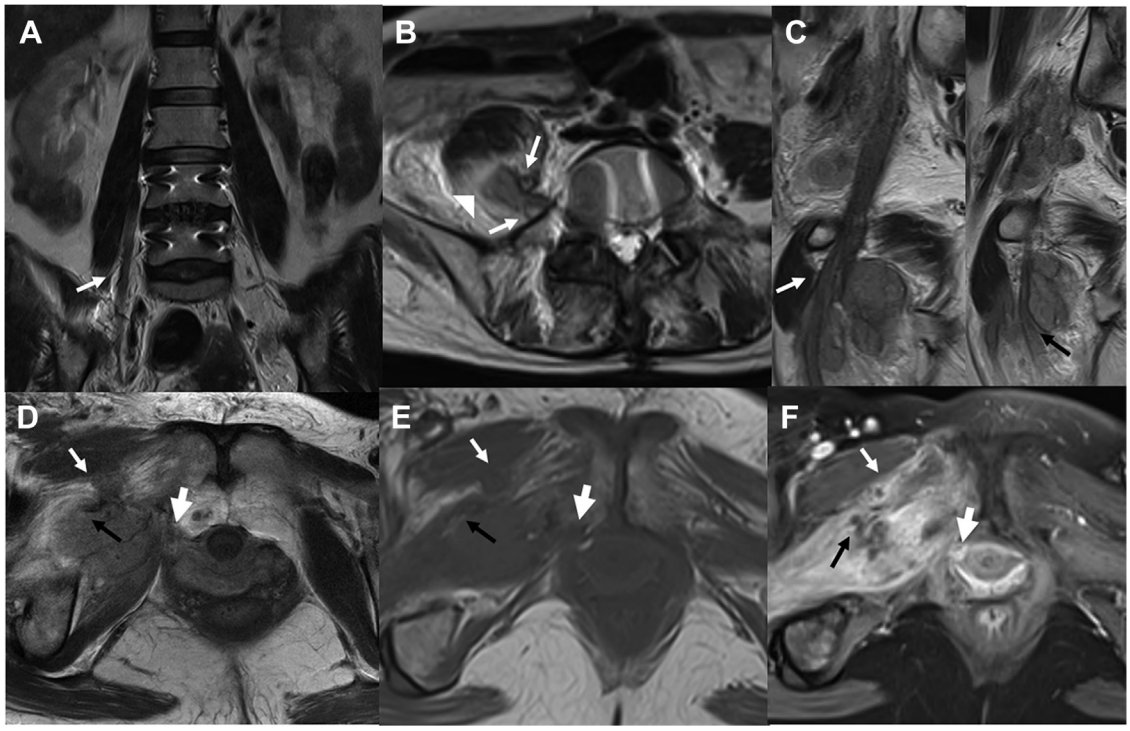

Figure 1. Magnetic resonance imaging of lumbar spine and pelvis. (**A**) Coronal T_2-weighted image shows asymmetric enlargement of right L3 nerve (*arrow*). (**B**) Axial T_2-weighted image shows perineural soft tissue mass (*thick arrow*) encasing right L3 and L4 nerve (*arrows*) with increased signal intensity and edema in the right psoas muscle. (**C**) Serial sagittal T_2-weighted images show diffuse thickening of anterior (*white arrow*) and posterior (*black arrow*) branches of obturator nerve with perineural masses in adductor muscles continuous with lumbar nerve. (**D**) Axial T_2-weighted image and (**E**) T_1-weighted image show enlargement and loss of the normal fascicular architecture of anterior (*white arrow*) and posterior (*black arrow*) branches of the obturator nerve with perineural masses in adductor muscles. The perineural masses are in contact with the right pelvic wall (*thick arrow*). (**F**) Axial fat-suppressed enhanced T_1-weighted image shows soft tissue mass with heterogenous enhancement in the right-sided wall of the vagina (*thick arrow*), with continuity to masses in the adductor muscles. The obturator nerve (*arrows*) adjacent to masses in the adductor muscles also shows heterogenous enhancement.

the obturator internus and adductor muscles was observed. The soft tissue masses in right adductor muscles surrounded anterior and posterior branches of the obturator nerve (Figure 1D–F). These masses showed similar signal intensity to those of the nerve lesions. They showed low-to-intermediate signal intensity on T_2-weighted images and heterogeneous enhancement. Conglomerated enlarged iliac and obturator lymph nodes near these perineural masses were seen, suggesting metastasis. Another lymphadenopathy was also present at para-aortic and internal iliac lymphatic chains. Based on these MRI findings, pelvic malignancy including vaginal cancer with perineural spread and lymph node metastasis, neurolymphoma, and atypical fungal infection were included in the final differential diagnosis.

Subsequently, ^{18}F-fluorodeoxyglucose-positron emission tomography/computed tomography (CT) demonstrated perineural spread, revealing increased uptake of L3 and L4 nerves and obturator nerve with perineural masses in the right psoas and adductor muscles (Figure 2). Soft tissue lesion in vagina and the adjacent right pelvic wall also showed increased uptake. Ultrasonography-guided biopsy on the right adductor muscle surrounding right obturator nerve and a punch biopsy on the vagina were performed. The 2 pathologic examinations showed moderately differentiated squamous cell carcinoma. The results are compatible with a diagnosis of moderately differentiated vaginal squamous cell carcinoma with perineural spread.

The patient was treated with 6 courses of chemotherapy with taxol and carboplatin and radiotherapy (30 Gy/10 fraction). One month later, the patient experienced a significant pain relief in the right thigh. At the 4-month follow-up, the patient was still alive, but the disease was aggravated with increased vaginal tumor size, numbers and size of metastatic lymph nodes, and progression of bone metastasis in the left pelvic bone. There was no significant interval change in diffuse perinueral spread of the disease.

DISCUSSION

Primary vaginal cancer is a rare disease, accounting for <2% of all pelvic malignancies.[7] Squamous cell carcinoma accounts for 80%–90% of primary vaginal cancer.[8] Primary vaginal squamous cell carcinoma tends to spread early by direct invasion of the bladder and the rectum, and by lymphatic invasion including pelvic or groin lymph node involvement.[8] However, at present, no cases on the perineural spread from vaginal squamous cell carcinoma have been reported.