Ureter and nerve root compression secondary to expansile fibrous dysplasia of the transverse process.

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**Corresponding Author:** Dr. Shuriz Hishmeh, MD

**Corresponding Author's Institution:** LI Spine Center

**First Author:** Shuriz Hishmeh, MD

**Order of Authors:** Shuriz Hishmeh, MD; Fernando Checo, MD; Alexandra Kondratyeva, DO

**Abstract:**
Urinary Retention Due To Transverse Process Fibrous Dysplasia

**Background Data:** Lytic lesions of the spine usually represent metastatic or infectious etiology. Benign tumors are commonly diagnosed as an incidental finding thus their true incidence is underreported.

**Purpose:** We describe a case report of monostotic fibrous dysplasia of a transverse process causing urinary symptoms.

**Study Design:** Case Study

**Methods:** We describe the management of a patient who presented with urinary retention and thigh numbness due to L3 transverse process lytic expansile lesion.

**Results:** After surgical intervention, the final pathologic diagnosis was fibrous dysplasia. The patient's urinary and radicular symptoms resolved with no documented recurrences.

**Conclusions:** Fibrous dysplasia is rarely seen in the spine and may mimic other pathological processes. The surgical and medical management of spinal fibrous dysplasia is described.
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Corresponding Author:

Shuriz Hishmeh, MD, PT
51 East Main Street
1st Floor
Smithtown, New York 11787
516-730-5042
Fax: 516.284.4243
NYSpineCenter@gmail.com

Fernando J Checo, MD
The Central Orthopaedic Group
651 Old Country Road, Suite 200
Plainview, NY 11021
516-681-8822
fernj75@aol.com

Alexandra Kondratyeva, DO
7600 Shore Front Parkway
Apt 4N
Arverne, New York 11692
646-287-3901
kondrat.alexandra@gmail.com
Ureter and nerve root compression secondary to expansile fibrous dysplasia of the transverse process.

Monostotic fibrous dysplasia involving the spine is rare. There have been only three cases reported of monostotic fibrous dysplasia isolated to the transverse process of the lumbar spine. We report a case of monostotic fibrous dysplasia of the lumbar spine involving the transverse process in a patient presenting with radiculopathy and urinary retention. We discuss the clinical symptoms, the CAT scan, Magnetic Resonance Imaging and histology findings along with a review of the literature.

**Case Report:**
Secondary tumors involving bony spinal column are relatively common, with post mortem studies indicating up to 70% of cancers patients having axial skeletal involvement (1,2). The vast majority of bone tumors are benign, however, due to the fact a majority of these tumors are clinically silent, their true incidence is hard to determine. Benign tumors are commonly diagnosed as an incidental finding thus their true incidence is underreported. More than 2000 primary bone and joint cancers and about 10,000 soft tissue Sarcomas are diagnosed in United States per year (3). The estimated incidence of benign primary tumors involving the spine accounts for about 1% of all primary skeletal tumors, and nearly 5% for malignant tumors (4,5,6).

The location within the axial skeleton also varies, with primary and secondary malignant osseous tumors have a predilection for the involving the anterior column an primary benign lesions affecting the posterior column (6,7,8,9). The most commonly encountered benign tumors affecting the spine include giant cell tumors, osteoid osteomas, osteoblastomas, and hemangiomas. It is estimated that Fibrous dysplasia accounts for approximately 7% of all benign bone lesions (10) and accounts for 1.4% of benign primary tumors affecting the spine (6).

This case report describes a patient who presented with an expansile lytic mass on a lumbar transverse process that was post operatively identified as monostotic fibrous dysplasia by pathology. To our knowledge there have been only three case studies describing monostotic fibrous dysplasia of lumbar spine in the transverse process (11,12). Expansile lytic bone tumors involving the transverse process is rare, but has been associated with pain and significant discomfort (10,11,12).

Fibrous dysplasia is a rare bone disorder caused by excessive proliferation of spindle cell fibrous tissues in bones. It is characterized by benign bone growths which can lead to local swelling, bony deformities and lytic conversion predisposing the bone to pathological fractures. Although this process can occur in cortical bone, it primarily affects the medullary space leading to enlargement and expansion from within the medullary space. Malignant transformation to osteosarcoma or fibrosarcoma can occur however its exceedingly rare (<0.5%) (13, 14).

There are types of fibrous dysplasia. The conventional isolated form, which may be monostotic or polyostotic was originally described in 1942 by Lichtenstein and Jaffe, and a polyostotic form associated with precocious puberty and café au lait spots (McCune-Albright syndrome).
Fibrous Dysplasia Causing Urinary Retention

Monostotic fibrous dysplasia or monostotic osteitis fibrosa comprises a majority of the cases of fibrous dysplasia (80%) (6,13,14,15).

Patient IK is an otherwise healthy 52 year old male, who over the previous six to eight months, was having increasing right flank pain, difficulty with urination, and right lower extremity L3 radiculopathy. A complete urological work up ensued with a series of invasive urological studies unable to explain his urinary symptoms. The patient ultimately had a CAT scan of his abdomen and pelvis. The CAT scan showed a mass of the right L3 transverse process (Figure A). This patient had a dedicated MRI of his lumbar spine showing an expansile lytic homogeneous mass to the patient’s right L3 transverse process leading to mass affect and compressing the exiting nerve roots, and presumably, his right ureter (Figure B). The patient has failed conservative management including physical therapy and NSAIDs and the right sided L3 radiculopathic pain was getting progressively worse, to the point where he was cutting back on his daily activities and requiring pain management. A discussion with the patient and urologist was had and the decision for surgical resection and biopsy was made.

On his history and review of system, patient IK denied any fevers, chills, headache, or change in weight, vision or hearing problems. He had no systemic symptoms except for six months of frequent urinary tract infections and difficulty with emptying his bladder and subsequent urinary retention. He denied any significant past medical history and denied any alcohol use nor any smoking history.

His physical examination, the only remarkable finding was tenderness along the iliopsoas and decreased fine and gross sensation to his right L3 dermatomal distribution. Motor examination was normal, no evidence of upper or lower motor neuron involvement. No signs of lower extremities erythema or infection were noted.

The patient was positioned prone on a Jackson Table onto a Wilson Frame. Fluoroscopy was used to localize the Right L3 transverse process. An incision was made over the right L3 transverse process and a Wiltse intramuscular approach was performed. After the right L3 transverse process was appreciated, a clamp was placed over the process and fluoroscopy was used to confirm our landmarks. After radiographic confirmation, we removed the soft tissue from the transverse process in all directions, including medially up to the pedicle. The inter-transverse ligament was detached from both the cephalad and caudal edges of the transverse process. We used a Woodson to perform sub-periosteal dissection to remove the soft tissue circumferentially. After dissection, we placed a cobb to protect the rostral and caudal soft tissue and used a Midas Rexx burr to amputate the lytic transverse process from its base. The transverse process was then removed enbloc (Figure C) and sent for pathological evaluation (Figures D,E).
Fibrous Dysplasia Causing Urinary Retention

Discussion:

Fibrous dysplasia causes an activating mutation in the gene that encodes the alpha subunit of the G protein slowing the differentiation of osteoblasts (2, 10). It presents in monostotic and polyostotic forms. Fibrous dysplasia is an expansile lytic intramedullary primary bone growth that has been found in all bones of the body including the skull, face, spine and extremities (2, 16, 17). Fibrous dysplasia is most often diagnosed by its radiographic appearance and biopsy, however, recent data suggest that DNA analysis may soon be able to diagnose this process (18).

The treatment varies from observation, biopsy to en bloc surgical resection. Clinical observation is warranted for asymptomatic or incidental findings of monostotic fibrous dysplasia as long as the risk for pathological fracture is low (2). Bisphosphonate therapy both oral and IV offers promising outcomes for the treatment of fibrous dysplasia with decrease in pain, improvement in function, and the radiographic findings (2, 19).

Progressive enlargement, recurrence and malignant transformation have been described in literature. Meredith and Healey reported reappearance of monostotic fibrous dysplasia affecting C2 extending through the fusion mass to involve a previously unaffected vertebra twenty years after the original C2 posterior elements excision with posterior spinal fusion C1 to C3 (14). In the literature the incidence of malignant transformation ranges from 0.4% to 4% (2). One case of malignant transformation in thoracic spine was reported by Fu and his colleagues (20). Therefore, complete removal of all affected bone is suggested (2, 11, 13, 14, 15, 17, 20).

The case of monostotic fibrous dysplasia presenting as an isolated expansile mass of the transverse process in lumbar spine has only been described a few times in the literature (11, 12, 13). Troop and Herring reported a case of monostotic fibrous dysplasia in the lumbar spine with involvement of the vertebral body and the posterior elements (12). Chow et al and Harris et al described the involvement of the transverse process of L4 (11, 13). Both reported lower back pain as clinical finding. Chow et al treatment consisted of excision that resulted in asymptomatic patient at 8-year follow-up, whereas, Harris et al chose observation. At 4-year follow-up patient’s lower back pain persisted.

According to the literature, the most common presenting symptom for patients with monostotic fibrous dysplasia of the spine is back pain localized to the lesion (15, 21). Meredith and Healey completed comprehensive review of fifty-four cases of monostotic fibrous dysplasia involving the spine where majority of symptoms included back pain, neck pain, sacral region pain, pathologic fracture, painful torticollis, progressive myelopathy, paresthesias of the foot, and only one case of radiculopathy involving thoracic vertebra (14).

We reported a case of monostotic fibrous dysplasia involving lumbar transverse process that presented with radiculopathy and urinary retention symptoms which has not been previously described. Due to progressive radiculopathy and urinary symptoms the treatment of choice was en bloc surgical resection. Post operatively complete resolution of symptoms was noted. This patient is now one year from his primary surgery and is doing well. His urinary symptoms and
his thigh radiculopathy have resolved. He will obtain serial imaging yearly due to the prevalence of recurrence.
References:

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