# The American Journal of Orthopedics

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

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## Abstract:

Considering their proximity to abdominal viscera, transverse process lesions may pose a diagnostic challenge. We present a case of fibrous dysplasia of a transverse process causing urinary retention, frequent urinary tract infections, and thigh numbness. This is the first reported case of a transverse process fibrous dysplasia lesion causing simultaneous urinary retention and neurologic symptoms. Clinicians may consider lesions of the lumbar transverse processes in patients presenting to orthopedic surgeons with urinary symptoms, especially when combined with neurologic symptoms. In these lesions, fibrous dysplasia should be within the differential. The diagnosis and a brief review of fibrous dysplasia is discussed.

## Response to Reviewers:

Reviewer #1:

"You mention that anterior spinal tumors are primarily malignant vs. posterior which is often benign. Perhaps focusing on the propensity for certain diagnoses in these particular locations is more appropriate than benign vs. malignant."

--we edited our discussion to include this topic. We discuss anterior vs posterior lesions and anatomic considerations.

"In the first line of the discussion, "More than 2,000 primary bone and joint cancers and about 10,000 soft tissue sarcomas are diagnosed in the United States each year, though a majority of these tumors are clinically silent and the true incidence is difficult to determine." These numbers are probably out of date.

--this line was removed and exchanged with a more up to date resource

"The description of the physical exam is somewhat sparse. Specifics related to the "L3 radiculopathy" symptoms would be useful."

--We expanded the physical exam and described his L3 radiculopathy

"Are there plain radiographs? These are often a useful diagnostic rest in FD."

--unfortunately, no plain radiographs or additional MRI cuts were available to use to
include in this report.

"Was a biopsy indicated before resection? Was a frozen section obtained during surgery? If one is unsure of diagnosis, a biopsy should be done even if planning wide resection."

--A frozen section biopsy was performed and our manuscript was edited to include this

- Anatomically the ureter is in front of the psoas muscle, far away from the transverse process. There does not seem to be edema in the psoas, so what is the mechanism for the urinary symptoms? Difficulty with urination should not be from a unilateral ureteral compression. Did he have hydronephrosis? Do you have imaging demonstrating compression of the ureter?

"Both low-power and high-power photomicrographs may not be necessary."

--we changed the figures and included only one photomicrograph

"Overall, this does show a unique presentation; however, it could be presented with an eye specifically towards diagnosis of FD, as well as the mechanics of the compression. I understand that the patient immediately got better, but I am unclear that we have explained this based on anatomy and physiology. Your discussion of FD itself is nice and concise, and mention of bisphosphonate therapy is quite appropriate."

--we appreciate the feedback. We updated our manuscript to present with an eye towards the diagnosis of fibrous dysplasia accordingly. We included relevant anatomy of the ureters in our discussion section as well

Reviewer 2:

"A discussion of the relevant anatomy and better imaging would be needed before consideration."

--we changed the manuscript to explain pertinent anatomy as well as anterior vs posterior lesions.

"Details of the prior urological evaluation and why this failed to uncover the problem earlier..."

--unfortunately, the patient only had a CT abdomen and pelvis to evaluate his symptoms, performed by his PCP. He was then referred to us after findings of an L3 transverse process lesion. We included relevant anatomy of the ureters in our discussion section as well.
May 13, 2015

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

Dear Reviewers:

Thank you for your consideration regarding this case report. We have identified a patient and diagnosis that we feel would benefit the medical community. We have not discovered this type of compression nor presentation in the literature.

This is the first reported case of a transverse process fibrous dysplasia causing urinary retention and neurologic symptoms simultaneously. Resection of the fibrous dysplasia resolved his urinary symptoms in its entirety. This manuscript has not been previously published in whole or in part and is not under review in any other medical journal. This manuscript has been read and approved by all authors. There were no conflicts of interest during this study and no funding provided. The authors have obtained the patient's informed consent for the case report.

Again, thank you for your considerations.

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This manuscript has been read and approved by all authors. There were no conflicts of interest during this study and no funding provided.

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Considering their proximity to abdominal viscera, transverse process lesions may pose a diagnostic challenge. We present a case of fibrous dysplasia of a transverse process causing urinary retention, frequent urinary tract infections, and thigh numbness. This is the first reported case of a transverse process fibrous dysplasia lesion causing simultaneous urinary retention and neurologic symptoms. Clinicians may consider lesions of the lumbar transverse processes in patients presenting to orthopedic surgeons with urinary symptoms, especially when combined with neurologic symptoms. In these lesions, fibrous dysplasia should be within the differential. The diagnosis and a brief review of fibrous dysplasia is discussed.
**Introduction:**

Fibrous dysplasia is a developmental abnormality caused by excessive proliferation of immature spindle-cell fibrous tissues in bones. It is characterized by benign bony 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 trabecular bone leading to enlargement and expansion from within the medullary space. Malignant transformation to osteosarcoma or fibrosarcoma can occur though exceedingly rare (<0.5%)\(^1,^2\). This case report describes a patient who presented with an expansile lytic mass in a lumbar transverse process that was postoperatively identified as monostotic fibrous dysplasia on pathology. Such lesions that involve the transverse processes are rare and have been associated with pain and significant discomfort\(^3,^4,^5\). This is the first reported case of a transverse process fibrous dysplasia causing urinary retention and neurologic symptoms simultaneously. The authors have obtained the patient's informed consent for the case report.

**Case Report:**

**History:**

A 52-year-old African-American male presented to us with six to eight months of increasing right flank pain, difficulty with urination, and right lower extremity pain in the area of his anterior thigh. He also complained of “buckling” of his thigh with ambulation. On review of systems, the patient denied any fevers, chills, headache, change in weight, vision or hearing problems. He had no systemic symptoms except for six months of frequent urinary tract infections and difficulty emptying his bladder resulting in urinary retention. He denied any significant past medical history and denied any alcohol use or tobacco history.

**Physical Examination:**
On physical examination, he was a well-appearing 52-year-old gentleman in no apparent distress. No signs of gross deformity, erythema, ecchymosis, or infection were noted upon examination of his lower extremities. His motor examination was within normal limits from L2 – S1. However, his sensation was decreased in the L3 distribution to both fine and gross sensation. Sensation was intact to the remaining nerve root distributions. Babinski’s test was negative both lower extremities and clonus was within physiologic limits. Examination of his gait was notable for quadriceps buckling with ambulation.

Radiographic Examination:

The patient initially presented to his primary care physician who evaluated his symptoms with a CT scan of his abdomen and pelvis. This revealed a mass of the right L3 transverse process (Figure 1). The patient was then referred to us for further management of this lesion. A dedicated MRI of his lumbar spine was subsequently performed, revealing an expansile, lytic, homogeneous mass in the patient’s right L3 transverse process. The mass demonstrated a significant mass effect compressing the exiting nerve roots and presumably his right ureter (Figure 2). A bone scan revealed monostotic disease. The patient had failed conservative management including physical therapy and anti-inflammatory medications. His right-sided radiculopathy was worsening and he complained that the pain was affecting his quality of life and limiting his abilities to perform his daily activities. Additionally, a pain management specialist was requested to better manage his pain. Considering progression of his condition, surgical management was discussed ultimately leading to a planned biopsy and resection of the mass.

Surgical Procedure:

The patient was taken into the operating room and positioned prone on a Jackson table with 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 identified, the soft tissue from the transverse process was retracted 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 elevator to perform sub-periosteal dissection to remove the soft tissue circumferentially. After dissection, we placed a Cobb elevator to protect the rostral and caudal soft tissue and used a high-speed burr to amputate the lytic transverse process from its base. The transverse process was then removed en bloc (Figure 3) and sent for frozen pathological evaluation (Figures 4). Following the diagnosis of a benign lesion, the wound was closed in layers.

Postoperative Course:
Complete resolution of both urinary and neurologic symptoms were immediately noted and up to 1 months postoperatively.

Discussion:
Primary bone tumors of the spine are rare, with a reported incidence of 2.5 - 8.5 per 100,000 people per year. 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. In contrast, secondary tumors involving the bony spinal column are relatively common. Postmortem studies indicate up to 70% of cancer patients demonstrate axial skeletal involvement. The most commonly encountered benign tumors affecting the spine include giant cell tumors, osteoid osteomas, osteoblastomas, and hemangiomas. Chordomas are frequently reported as the most common malignant primary spine neoplasms. Of all primary benign bone lesions, fibrous dysplasia accounts for approximately 1.4%.
Primary and secondary malignant osseous tumors have a predilection for the anterior column and primary benign lesions usually affect the posterior column\textsuperscript{1,12,13,14}. Because of the greater blood supply and more direct communication with the viscera via Batson’s plexus, the anterior column is most likely to be seeded by metastatic disease. Similarly, hemangiomas and multiple myeloma are typically located in the anterior column most likely because of the more abundant blood supply there. Chordomas are also found in this cancellous anterior column. Osteoid osteoma, osteoblastoma, and aneurysmal bone cysts are found within the more cortical architecture of posterior elements. This patient’s lesion within the transverse process elevates confidence in a benign lesion.

The conventional, isolated form of fibrous dysplasia was originally described in 1942 by Lichtenstein and Jaffe. They described 15 cases of benign “non-osteogenic fibromas” near the ends of long bones in young patients. Monostotic fibrous dysplasia comprises the majority of these cases, approximately 80\%\textsuperscript{1,2,8,15}. Fibrous dysplasia may also present as part of McCune-Albright syndrome in which case it is associated with precocious puberty and café au lait spots. Less commonly, they are associated with intramuscular myxomas as in Mazabraud syndrome. The lesions in these syndromes are typically polyostotic. In all forms, fibrous dysplasia develops from an activating mutation in the gene that encodes the alpha subunit of the G protein on chromosome 20q13, activating cyclic adenylate cyclase and slowing the differentiation of osteoblasts \textsuperscript{38}.

With regard to presentation, fibrous dysplasia is usually asymptomatic and discovered incidentally. The literature reports the most common presenting symptom for patients with monostotic fibrous dysplasia of the spine is back pain localized to the lesion\textsuperscript{15}. Meredith and Healey completed a comprehensive review of fifty-four cases of monostotic fibrous dysplasia involving the spine
where the 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. In normal anatomy, the ureter lies within retroperitoneal fat anterior to the psoas muscle and L2 – L5 transverse processes and is normally mobile. This becomes clinically significant in lean patients as the ureter becomes closer to the spine. There are several reports of iatrogenic ureter injury in lumbar disc surgery. Normal variants, including medialization towards the spine, may predispose the ureters to injury, iatrogenic or otherwise. In fact, medialization of the ureters occurs commonly in African-American males and usually involves the right side, which may have occurred in this African-American patient.

Fibrous dysplasia is most often diagnosed by its radiographic appearance or biopsy. However, recent data suggest that DNA analysis may soon be able to diagnose this process. Imaging typically reveals expansile, central lytic lesions within the medullary cavity. Pathology demonstrates dense fibroblasts around immature woven bone, commonly referred to as “Chinese lettering”. The treatment varies from observation 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. Bisphosphonate therapy, both oral and IV, offers promising outcomes for the treatment of fibrous dysplasia with improvement in pain and function, as well as the radiographic findings. Management of monostotic fibrous dysplasia presenting as an isolated expansile mass of the transverse process in lumbar spine has rarely been described. Troop and Herring reported a case of monostotic fibrous dysplasia in the lumbar spine with involvement of the vertebral body and the posterior elements. Chow et al and Harris et al described the involvement of the transverse process of L4. Chow et al’s treatment consisted of excision that
resulted in asymptomatic patient at 8-year follow-up, whereas Harris et al chose observation. In the latter study, the patient’s lower back pain persisted at 4-year follow-up.

Progressive enlargement, recurrence, and malignant transformation have all been described. Meredith and Healey reported the 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. In the literature, the incidence of malignant transformation ranges from 0.4% to 4%. One case of malignant transformation in thoracic spine was reported by Fu and his colleagues. Therefore, complete removal of all affected bone is recommended.

We present an unusual condition with complete resolution of symptoms following surgical resection. Several points may be considered from this report. Fibrous dysplasia lesions have been found in all bones of the body including the skull, face, and extremities but monostotic fibrous dysplasia involving the spine is rare. Furthermore, there are no other reports of these lesions causing simultaneous nerve compression and urologic symptoms. Considering anatomy, clinicians may consider lesions of the lumbar transverse process in patients presenting to orthopedic surgeons with urinary symptoms, especially when combined with neurologic symptoms. In these lesions, fibrous dysplasia should be within the differential diagnosis. Clinicians should also recognize that complete resolution of symptoms has been reported with wide resection of these lesions.
References:
7. Schuster JM, Grady MS: Medical management and adjuvant therapies in spinal metastatic disease. Neurosurg Focus 2001;11:E3


**Figure Legend:**

**Figure (1)** CT Scan demonstrating an expansile lytic lesion involving the right L3 transverse process with thinning of the cortex. This mass is posterior to the right psoas muscle. It measures 2.3 x 1.7 x 1.5 cm.

**Figure (2)** MRI showing a discrete well defined right pedicle lesion measuring 2.7 x 1.7 cm with bone marrow edema.

**Figure (3)** Gross specimen

**Figure (4)** Low magnification showing an irregular collection of small pieces of immature woven trabeculae surrounded by a fibrous-appearing stroma composed of spindle cells (hematoxylin and eosin, x100).
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Figure (3) Gross Specimen
Figure (4) Low magnification showing an irregular collection of small pieces of immature woven trabeculae surrounded by a fibrous-appearing stroma composed of spindle cells (hematoxylin and eosin, x100).
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