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Chordoma, An Incidental Finding in A Patient with Low Back Pain and Urinary Incontinence: A Case Report

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Abstract

Purpose: A chordoma is a rare mesenchymal tumour that occurs in the midline from clivus to sacrum.It is thought to originate from transformed remnants of notochord and has a prediletion for axial skeleton. A chordoma biological behavior is characterized by a slow aggressive local growth with a low tendency in metastasizing to distant sites including the lungs, bone, soft tissues, lymph nodes, liver and skin.

Materials and Methods: We report an incidental finding of a case of lumbar chordoma in a 48-year old man who presented with a 7-year history of low back pain and urinary incontinence after a fall from a two storey building. At presentation, we made a diagnosis of traumatic neurogenic bladder but magnetic resonance imaging scan of lumbosacral spine revealed retropulsed collapsed 1st lumbar vertebra with cord compression.

Findings: Spinal decompression done by the removal of the caseous material which was sent to histology revealed the diagnosis of chordoma. The epidemiology, clinical evaluation, diagnostic imaging and treatment modalities have been discussed with review of existing literature.

Implications to Theory, Practice and Policy: It is important to have high index of suspicion of chordoma on any patient who presents with low back pain and urinary incontinence.

Keywords: *Chordoma, Incidental, Pain, Incontinence, Compression, Lumbosacral*

JELCodes: I30, I30, I30, I30, I30, I30, I30

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INTRODUCTION

Chordomas are rare mesenchymal tumours occurring exclusively in the midline from clivus to sacrum. They involve both ends of the axial skeleton and present as destructive bone lesions with a large soft tissue mass. Chordomas are thought to arise from transformed remnants of notochord and have a prediletion for the axial skeleton, with the most common sites being the sacrum, skull base and spine. Due to its ectodermal origin, chordoma is not properly a sarcoma even if it has clinically retained and classified as such being a primary tumour of bone.[1]

The sacrum represents the most common anatomical site of origin accounting for 50-60% of all cases followed by the skull base region (spheno-occipital/nasal) 25-35% of cases and thoracolumbar vertebrae approximately 5% of cases.[2]

Chordoma biological behaviour is characterized by a generally slow aggressive local growth with a low to late tendency in metastasizing to distant sites including the lungs, bone, soft tissues, lymph nodes, liver and skin[2.] Although histologically considered to be a low grade neoplasm, chordomas are highly recurrent, making their clinical progression very similar to that of malignant tumours[3].

Chordomas of the mobile spine have distinct treatment considerations from tumours of the clivus and sacrum. A soft tissue component is often present and may invade the paravertebral musculature. Although commonly described as benign, chordomas have malignant potential, and nodal metastases and haematogenous dissemination may develop in more than one-third of patients[4].

Case Report

A case of 48 year old man admitted on account of 7year history of urinary incontinence. The patient was said to fell down from a two storey building while roofing the building. He was unable to tell which part of the body he fell down with, though no history of loss of consciousness or bleeding from craniofacial orifices. He was unable to move both lower limbs but no loss of sensation. He was immediately transported to nearby private hospital on the back of a car, where he was resuscitated and later catheterized on account of difficulty in micturition. He was referred to university of Port Harcourt Teaching Hospital, Port Harcourt where he was managed subsequently. He noticed that he was incontinent of urine three week after the removal of urethral catheter. There was associated weak erection, occasional constipation but no faecal incontinence.

He has been having recurrent low back pain, burning in nature, occasionally radiates to back of the thigh and leg, aggravated by standing and walking for a period of time. Relieved by sitting and lying down, no claudication. He has been on urethral catheter which was changed two weekly for urinary incontinence. He decided to visit our centre for further evaluation and care. He has no co-mobility or history of surgery in the past, a carpenter Stopped cigarette smoking about ten years ago. Examination revealed a young man in no distress with normal vital signs. Neurological examination revealed normal sensory modalities but power of grade 4 on left ankle dorsiflexors and plantarflexors. Other muscle bulks on the lower limbs have power of 5.Tone and reflexes were normal.

There was tenderness on the lumbosacral region. Genitourinary system revealed a two- way Foley urethral catheter in situ with a spigot mechanism. The catheter with spigot mechanism was used because the patient was having urinary incontinence, hence, he could void only when the spigot was removed. Digital rectal examination revealed lax anal sphincteric tone with empty rectum. Diagnosis of traumatic neurogenic bladder was made. Lumbosacral x ray showed wedge compression fracture 1st lumbar vertebra whereas magnetic resonance imaging

of the lumbosacral spine revealed retropulsed collapsed 1st lumbar vertebra with cord compression. Urinalysis, serum electrolyte urea and creatinine were normal. Full blood count showed neutrophilia with total white blood cell within normal limit and elevated erythrocyte sedimentation rate. Mantoux test result was 15mm.

Research Question

Can chordoma be the cause of recurrent low back pain and urinary incontinence in a 48year man?

Statement of Problem

Low back pain and urinary incontinence are debilitating symptoms in any adult, it is therefore essential that their aetiology should be elucidated to know the right treatment options to be used in such patient management.

Method

He had anterior decompression of 1st lumbar vertebra and spinal fusion with tricortical bone graft, done under general anaethesia with cuffed endotracheal intubation, antibiotics prophylaxis given at induction. Retroperitoneal approach through left subcostal incision. Anterior wedge collapse of the body of 1st lumbar vertebra with caseous necrosis of the right lateral part, spinal stenosis with cord compression noted as intraoperative findings.

Decompression achieved by the removal of the caseous necrotic material and bone biopsy specimen taken for histology. Primary haemorrhage was significant which was arrested. Wound irrigated with saline. Tricortical bone graft collected from the left anterior iliac bone and used to stabilize the 1st lumbar vertebra. Suction drain inserted and wound closed in layers. He received three units of blood intra operatively. The procedure was well tolerated by the patient.

Postoperatively, parenteral antibiotics, analgesics and anti-thrombotics were commenced. Bladder drainage was continued. Anti-kochs therapy was also commenced after the return of bowel sounds. Suction drain was removed on the 3rd day post operatively. Sutures were removed at the 14th day post operatively. Oral muscle relaxant was commenced, thoraco-lumbar jacket applied. He was discharge on the 25th day post operatively.

First post visit, four weeks after discharge, the low back pain had reduced significantly, he was ambulating well, still on spigotted urethral catheter. The thoracolumber jacket was removed and changed to lumber corset. Histology report revealed chordoma of lumbar spine.

Discussion

Chordoma is a rare cancer that accounts for 1-4% of all bone malignancies.[5] chordomas occur at any age but are usually seen in adults (30-60 years). Those located in the spheno-occipital region most commonly occur in patients 20-40 years of age, whereas, sacrococcygeal chordomas are typically seen in a slightly older age group (peak 40-60 years). Population based studies using surveillance, epidemiology, and end result (SEER) databases suggest an incidence of chordoma of 0.08 per 100,000, with predominance in men and peak incidence between50 – 60 years of age[6]. For unknown reason, it is rare in African Americans. Chordomas have very low incidence in patients younger than 40 years, and rarely affect children and adolescents (<5% of all chordoma cases).[6,7]

It develops in males most often than females and, for unknown reasons, is rare in African Americans. In the united states, the annual incidence of chordoma is approximately one in one million (300 new patients each year)[8].

Chordomas were first characterized microscopically by Virchow in 1857[9]. He described unique, intracellular bubble-like vacuoles that he referred to as physaliferous, which in a distinguishing feature if not pathognomonic for chordoma[9]. Virchow hypothesized that chordomas were derived from cartilage; however, more contemporary evidence suggests that they are derived from undifferentiated remnants that reside within the vertebral bodies and throughout the axial skeleton[9].

A small number of families have been reported in which multiple relatives have been affected by chordomas. In four of these families duplication of the brachyury gene was found to be responsible for causing chordoma[10]. Brachyury regulates several compelling stem-cell genes and has recently been implicated in promoting epithelial mesenchymal transition in other human carcinomas.[11].

A possible association with tuberous sclerosis complex (TSC1 or TSC2) has been suggested[12]

Chordomas are indolent and slow-growing, therefore, they are often clinically silent until the late stages of disease. Symptoms are generally of long duration due to slow growth of lesion and delay in diagnosis. They are usually nonspecific, depending on location of tumour.

Skull-base chordomas often grow in the clivus and present with cranial nerve palsies. Depending on their size and involvement of the sella, endocrinopathy can also occur[13]. Other rare presentations include epistaxis and intracranial haemorrhage[14,15].

Cervical spine chordoma presents with: airway obstruction, hoarseness, dysphagia, bleeding, oropharyngeal mass. Sacral chordomas present with: vague low back pain, constipation, low extremity radiculopathy, palpable mass, bowels and bladder dysfunction resulting from local organ mass effects or autonomic dysfunction. Autonomic dysfunction can result as the tumour compresses the adjacent sympathetic or parasympathetic outflow.

Chordomas of the mobile spine and sacrum can present with localize deep pain or radiculopathies related to the spinal level at which they occur[16,17].

Most saccrococcygeal chordomas involve the fourth and fifth sacral vertebrae, and although large tumours can protrude anteriorly into the pelvis, invasion into pelvic structures is often limited by presacral fascia[18].

Several imaging tests may be required to identify a chordoma; including x –rays, computed tomography (CT) scans, fracture magnetic resonance imaging (MRI), and bone scan.

Chordomas, themselves do not show up well, but the bone damage may be seen on x-rays. In plain films; lesion may be missed, changes are generally subtle.

Findings may be masked by overlying at abdominal bowel gas on AP of pelvis. Also osteolytic sacral lesions may be overlooked on plain radiographs.

MRI and CT scan have complementary roles in tumour evaluation. CT evaluation is needed to assess the degree of bone involvement or destruction and to detect patterns of calcifications within the lesion.

MRI provides excellent 3-dimesional analysis of the posterior fossa (especially the brains stem), sella turcica, cavernous sinuses and middle cranial fossa. MRI depicts calcifications and the precise involvement of skull base osteolysis less well than CT, especially for skull base foramina. Centrally located, well circumscribed, destructive lytic lesion, sometimes with marginal sclerosis. Expansile soft tissue mass (usually hyper- attenuating relative to the adjacent brain, however, in homogenous areas may be seen due to cystic necrosis or

haemorrhage, the soft tissue mass is often disproportionately large relative to the bony destruction. Irregular intratumoral calcifications with moderate to marked enhancement.

In MRI, T1 – Intermediate to low signal intensity-Small foci of hyperintensity (intratumoral haemorrhage or mucus pool). T2 – most exhibit very high signal. T1C (Gd); hetergenous enhancement with a honey comb appearance corresponding to low T1 signal areas within the tumour. GE (Gradient Echo) confirms haemorrhage if present with blooming. Bone scan; there may be normal or decreased uptake.

Treatment of chordomas is very difficult. This is mainly because these tumours are near the brain and spinal cord. Various treatment modalities have been adopted for treatment of chordomas such as:

Surgery is the preferred treatment option where possible. In the 1970s, Stener and Gunterberg[19] first introduced the idea of wide en-bloc surgical resection for the treatment of sacral tumours. Since then, en-block excision has remained a central tenant in the surgical management of scaral chordoma.

With the advent of more aggressive surgery and wider surgical margins, local control of disease recurrence has substantially improved for chordomas of the sacrum, spine and skull base.

Despite major advances in surgical interventions, total en-bloc resection is attainable in roughly 50% of sacral chordomas, with much lower rates for chordomas of the spine and skull base. The use of radio-therapy as primary or adjuvant treatment for chordoma is debated. Radiotherapy alone has proven to be ineffective when coupled with debulking or palliative therapy[20].

Advances in radiation technology with the introduction of hadrons have led to higher dose of radiation being delivered to the target volume, with minimum injury to the surrounding tissue and improved radiobiological effect[21].

Anthracyclne, cisplatin, alkylating agents, and camptothecin have been reported to affect chordomas, and some case reports have suggested sensitivity of one of the histological variants – dedifferentiated chordoma[22].Although surgical resection has been the first line of treatment in feasible scenarios with radiotherapy offered for recurrent case. Some advocate for combination of radiotherapy and complete or subtotal surgical resection for selected patient.

The complications associated with chordoma depends on the site, size and grade; For clival chordomas; cranial nerve palsies, epistaxis, intracranial haemorrhage. Cervical spine chordoma, airway obstruction, dysphagia. Sacral chordomas; low back pain, low extremity radiculopathy, bowel and bladder dysfunctions.

The main complications of treatment are: postoperative cerebrospinal fluid leakage in clival chordomas, intraoperative arterial injury, new onset cranial nerve deficits, perioperative death occurs more often in patients who had undergone previous treatment, bowel and bladder dysfunction in resection of lumbar and sacrococcygeal chordomas. Intracranial haemorrhage in clival chordomas.

The multidisciplinary approach continues throughout the patient admission including general surgical oncologists, thoracic surgeons, ear, nose and throat specialists (ENT); and reconstructive surgeons; neurological surgeons, Orthopaedic oncology surgeons, radiation oncologists, and clinical nurse specialists; social workers, physical therapists working as an outcome for the individual patient.

Frequent follow-up is required because of high rate of recurrence of these tumours. The average interval to recurrence is 2.27 years for radically resected tumours, 2.1 years for subtotal

resection followed by radiation therapy and 8 months for subtotal excision without, adjuvant therapy. The interval of follow-up, including repeat MRI or CT scans, depends on the completeness of the resection[10].

Chordomas are relatively benign appearing neoplasms, however, because of their tendency to erode bone and invade soft tissues, they usually display malignant behaviour.

"The 5-year survival rate is estimated to be 76.7% and the 10-year survival is 59.1%[22]". Factors that may improve prognosis are young age, complete resection and the addition of radiation therapy in completely resected tumours[23].

Conclusion

The diagnosis of lumbar chordoma remains uncommon, however, it should be considered in patients with dull low back pain especially associated with change in urinary and bowel habits. Lumbar chordoma is curable with surgery and early diagnosis may lead to the preservation of bowel, urinary and sexual functions.

Recommendation

The chordoma as rare as it is should be considered as a possible diagnosis in a patient with low back pain and urinary incontinence. Therefore, spinal surgeons should have high index of suspicion in any patient who presented with low back pain and urinary incontinence and ensure that sample specimen be collected in any patient undergoing any spinal operative procedure for low back pain.

Conflict of Interest: Nil.

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