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Case Report

Fibrous dysplasia in axis treated with vertebroplasty

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Abstract

Vertebroplasty of the axis is a challenging procedure, and little is known about its therapeutic outcome. Cervical fibrous dysplasia with a distinct cyst is a rare entity and few cases have been reported in the literature. A 55-year-old man with fibrous dysplasia of axis presented with severe neck pain and left arm since six months. Computed tomography and magnetic resonance imaging revealed an expansile, destructive lesion involving the axis, and no spinal cord. He was submitted to retropharyngeal surgery and the lesion was fulled by vertebroplasty. Microscopic examination was consistent with the diagnosis of monostotic fibrous dysplasia. After the surgery no recurrence was observed. The patient had remarkable improvement in clinical relief of neck pain at 1-year follow-up. Although there are descriptions of vertebral fibrous dysplasia, this is the 13th case of monostotic fibrous dysplasia of the cervical spine, and the 3rd case of the axis described in the literature. The unique case who had treated with ope vertebroplasty.

Key words: Axis, fibrous dysplasia, spine, vertebroplasty

INTRODUCTION

Fibrous dysplasia (FD) is an uncommon benign fibroosseous abnormality of cervical spine, of unknown aetiology and equal sex incidence.[1] In certain individual with genetics predisposition multiple bones can be affected especially in syndromes including McCune–Albright. Surgery is performed when the patient has persistent pain, neurologic impairment, vertebral collapse, instability, and/or cord compression.[2] Vertebroplasty can be performed in the axis either by a direct transoral[3-5] or an percutaneus anterolateral approach;[6] from a technical point of view, either approach is much more difficult compared with that of transpedicular percutaneous vertebroplasty in the thoracic and lumbar spine because of potential dangerous complications related to the spinal cord, jugular vein, cranial nerves, carotid artery, and vertebral artery. The treatment of percutaneous vertebroplasty for the fibrous dysplasia in the axis is reported before only one once in the literature.[7]

To our knowledge, no clinical or case studies have evaluated the feasibility, safety, and efficacy of vertebroplasty for the axis. The aim of the present study was to evaluate the risk and benefits of vertebroplasty for the treatment of axis fibrous dysplasia using an retropharyngeal approach.

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CASE REPORT

A 55-year-old male patient presented with pain in the back of the neck. He had been getting vague discomfort along the cervical spine since one year. Since two months he noticed tingling sensation on the left side of the body and fingers on bending or turning the neck to the left. No definite history of trauma was noted. Patient was not diabetic or hypertensive. Patient gave no history of sistemic illness. On examination the (O/E) patient was conscious, responsive, oriented, right-handed. Speech was normal. No cranial nerve deficits. No motor deficits. No evidence

of wasting of muscles. No fasciculations. Deep tendon jerks were symmetrical. Plantars were equivocal on both sides. Visceral reflexes were normal. Sensory system examination revealed dysasthesia over the suboccipital region. Examination of the cervical spine did not present any abnormality. No abnormal mass was palpated over the paraspinal region or over the spinous process. Left lateral flexion of the neck was restricted due to pain going down the left side of the body. Patient was investigated with direct cervical graphy, computed tomography (CT) and magnetic resonance imaging (MRI) scans of cervical spine. Direct lateral cervical spine showed lytic appearence of the axis. In the CT of cervical spine an irregular bony growth was seen along entire axis and expansil vertebral body of the axis without compressing the spinal cord. CT showed a ground-glass appearance [Figure 1]. MRI scan of the cervical spine [Figure 2]. A bony outgrowth was seen arising from the entire of axis projecting and without compressing the spinal cord at that level. Cervical spinal cord was appeared free. The cortex was thinned and expanded, and encroached upon the left C2 foramen transversarium and spinal canal. MRI demonstrated hypointensity on both T1 and T2, with minimal contrast enhancement. A provisional diagnosis of fibrous dysplasia/plasmcytoma was made by the radiologist. He was operated by left retropharyngeal approach. Axis was seperated by right side. The tumor was curetted [Figure 3], and cement was filled to be starting [Figure 4] tumor was sent for histopathological examination. Vertebroplasty was performed into the cavity by

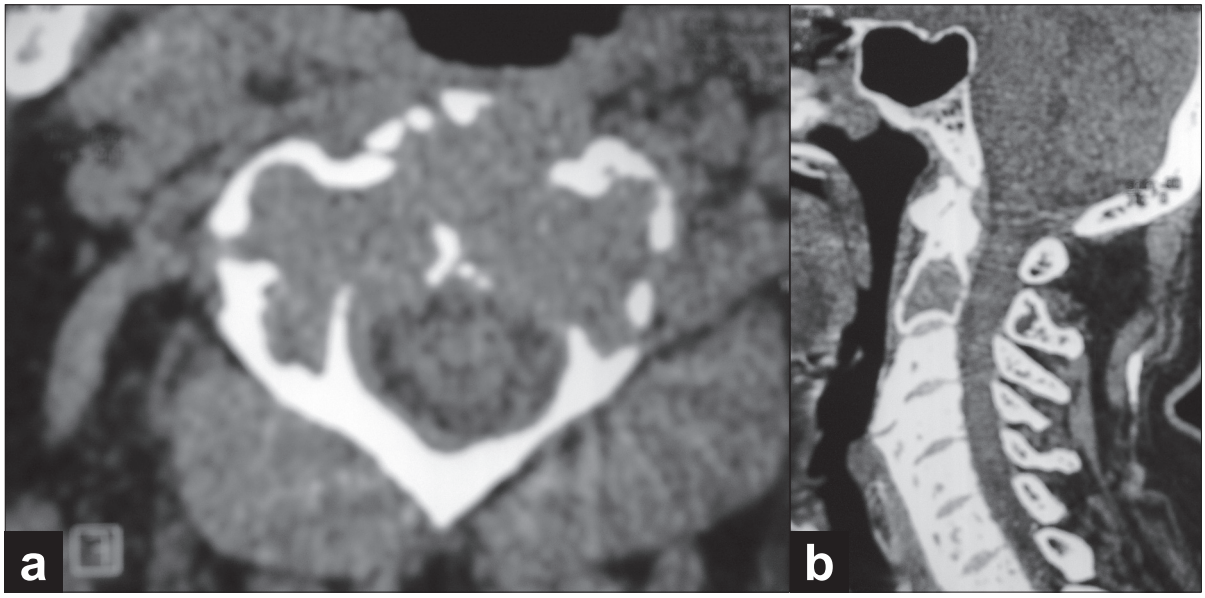

Figure 1: Computed tomography scan of the cervical spine (a; transverse cut, b; sagittal cut) showing fibrous dysplasia axis (ground-glass appearance)

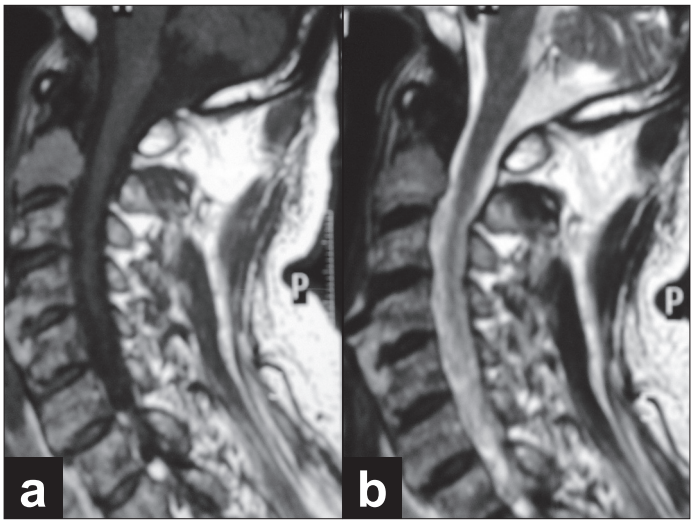

Figure 2: Magnetic resonance imaging scan of the cervical spine (sagittal section) showing the dysplastic of axis body. MRI demonstrated hypointensity on both T1 [a]and T2 [b], with minimal contrast enhancement

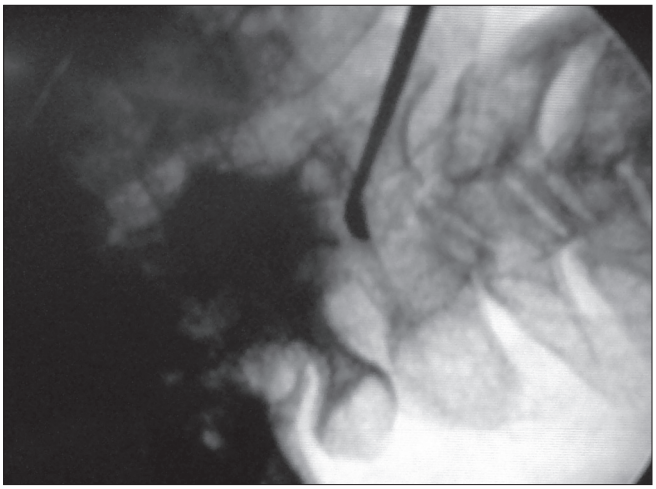

Figure 3: The tumor was curetted in the peroperative period

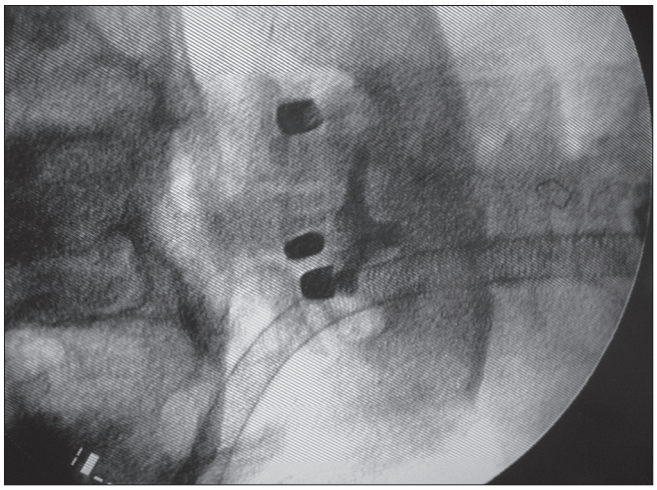

Figure 4: The cement beginning to fill in the peroperative period

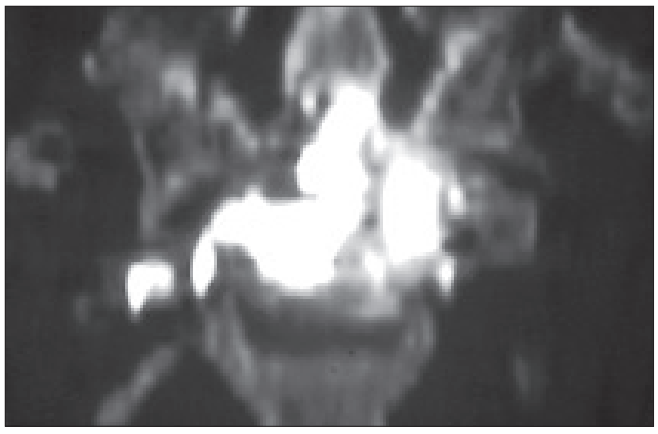

Figure 5: Coronal CT obtained after placement of the needle shows its oblique trajectory and the approach to axis. Note that the needle is placed at the central part of the axis

open surgical method. Only 3 cc polymethylmethacrylate was injected into the body of axis [Figure 5]. No any complication such as cement leakage was observed. Postoperatively the patient made a good recovery. Bisphosphonate therapy was offered to patient for the prophylaxis.

DISCUSSION

The term fibrous dysplasia was used first by Lichtenstein[8] in 1938 to designate a developmental anomaly of unknown etiology characterized by fibrous tissue replacement of medullary cavity of bones. Any bone in the body can be affected. The involvement can be either monostotic or polystotic. Monostotic fibrous dysplasia of the spine is a rare entity.[7,9] The management has been reported in many patients who had pathologic fracture, neurologic deficits, persistent neck pain with or without deformity[9-15] Surgery is indicated for confirmatory biopsy, correction of deformity, prevention of pathologic fracture, and/or eradication of symptomatic lesions. The use of cortical grafts is preferred over cancellous grafts or bone-graft substitutes because of the superior physical qualities of remodeled cortical bone. The natural history and ideal treatment for this condition remain poorly understood. Surgical approach is advocated to be tailored to the involvement of the dysplastic tissues. Many researchers offer both posterior and anterior stabilization[11-13,15] Curettage with bone graft placement can be performed but the dysplastic tissues have been found to extend into the grafted materials.[16,17] The risks involved with surgery of cervical spine fibrous dysplasia can be potentially high. The bone is fragile as the cortex may be eggshell skinny.[9,11,14,16] But, vertebroplasty does not use conventional surgery. We present a potential treatment of vertebral fibrous dysplasia with vertebroplasty. This technique has been used for the treatment of osteoporotic vetebral collapse, vertebral angiomas, and malignant tumors.[1,7,18-21] Vertebroplasty provides mechanical support and pain control. We project that the complication rate for fibrous dysplasia treatment would be less than or comparable to that of angioma, which is approximately 2–5%.[19,20] The main complication is radiculopathy from cement leakage into the epidural or foraminal veins.[1,7] Most investigators have spoken that the presence of symptoms from spinal cord compression is a contraindication to the performance of vertebroplasty.[20] We performed open vertebroplasty due to cement leakage into the spinal canal. On the basis of our result, we believe that open vertebroplasty by an retropharyngeal route is an effective procedure.

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