Spinal Compression by Brown Tumor in Two Patients With Chronic Kidney Allograft Failure on Maintenance Hemodialysis

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Brown tumors with non-neoplastic process are noticed in patients with end-stage renal disease suffering from a severe form of secondary hyperparathyroidism. Herein, we report a patient with chronic kidney allograft failure returned back to hemodialysis who experienced manifestations of cauda equina compression secondary to a lumbar brown tumor. Also, we had another patient on hemodialysis with a demineralized lesion affecting the cervical vertebrae. Although brown tumor is a rare complication, these two cases highlighted the importance of neurological symptoms in uremic patients. Spinal decompression surgery, in order to alleviate pressure on neurological structures, together with subtotal parathyroidectomy, were highly indicated.

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Case Report

INTRODUCTION

Brown tumors are erosive bony lesions caused by increased osteoclastic activity and peritrabecular fibrosis (*osteitis fibrosa*) secondary to phosphate retention and impaired vitamin D synthesis. These cystic lesions of the bones may appear in any of the bones, but are frequently found in the pelvic bones, facial bones, ribs, mandible, and less commonly affecting, the vertebrae.[1] To the best of our knowledge, 6 cases of thoracic and 2 cases of cervical brown tumors have been diagnosed by 2005.[2-9] The age range of these patients was 24 to 69 years. Six patients had a dialysis period ranged from zero to 11 years, and 2 were diagnosed after graft rejection or failure (1 thoracic and 1 cervical).[3,8] Herein, we report 2 cases of brown tumors affecting the spine in 2 patients with chronic kidney allograft failure who returned back to dialysis for 3 years.

CASE REPORT

Case 1

The first case was a 19-year-old man who had received his kidney allograft from his mother in 2001. The original kidney disease was idiopathic fibrocellular crescentic glomerulonephritis that was not responding to steroid pulse. After 3 months of hemodialysis with acceptable parathyroid hormone, he underwent kidney transplantation. The immunosuppression protocol consisted of antithymocyte globulin induction, and then the patient was maintained on steroid, cyclosporine, and azathioprine. An acute rejection episode occurred that was treated by pulse steroid with partial improvement. Few weeks later, he suffered bilateral avascular hip necrosis for which bilateral hip decompression was performed. Chronic deterioration of the graft developed as a result of chronic allograft nephropathy, and subsequently, he returned back to regular hemodialysis.

He was doing well on hemodialysis till the 3rd year, when he presented with persistent low backache, which was associated with numbness and tingling of both lower limbs, especially on walking. Physical examination revealed weakness

of the left foot flexors, absence of the left ankle reflex, with a positive bilateral Lasegue and Lhermitte signs. Sphincter tone and sensation were all normal. Laboratory findings revealed elevated serum alkaline phosphatase (405 IU/L), parathyroid hormone (985 pg/mL), and phosphate (6.9 mg/dL); however, serum calcium level was (9.5 mg/dL). Lumbosacral plain radiography showed demineralized lesion affecting the 3rd lumber vertebra. Further radiological assessment by magnetic resonance imaging demonstrated bone infiltration of that vertebra with large paravertebral and intraspinal soft tissue masses with neural compression (Figure 1). Moreover, bone isotope scan was performed to exclude metastasis, while parathyroid adenomas were documented on ultrasonography.

Surgical removal of the tumor was performed with dural sheet reconstruction after nerve root dissection. Pathologic examination of the excised mass revealed granulation tissue with fibroblasts, histiocytic cells, few plasma cells, and lymphocytes. Numerous osteoclastic giant cells and osteoid-rich bone fragments were present, with microfoci of hemosiderin deposition. Parathyroid adenoma was removed later by subtotal parathyroidectomy that resulted in normalization of the parathyroid hormone level. High-dose calcium (both oral and intravenous) was administered for a few months to maintain the serum calcium levels within the normal range. Following surgical intervention, clinical improvement was achieved after 3 weeks with complete mineralization of the spine without nerve defects.

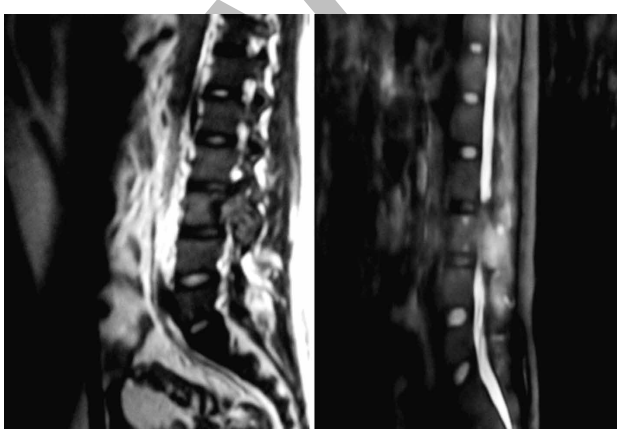

Figure 1. Magnetic resonance imaging demonstrated bone infiltration of that vertebra with large paravertebral and intraspinal soft tissue masses and neural compression.

Case 2

The second case was 25-year-old woman who received her kidney allograft from her mother in 2002. She had received basiliximab induction, followed by steroid, cyclosporine, and azathioprine as maintenance immunosuppressive therapy. During the follow-up period an acute rejection episode developed that was treated by pulse steroid with partial improvement; however, chronic deterioration of the graft as a result of chronic allograft nephropathy led to returning back to regular hemodialysis.

After 2 years of hemodialysis, she presented with persistent neck pain, which was associated with numbness and tingling of both lower limbs, especially on walking, and root pains in both upper limbs. Physical examination revealed muscle weakness and hypotonia of both upper limbs. Laboratory studies revealed elevated serum alkaline phosphatase (322 IU/L), parathyroid hormone (2600 pg/mL), and phosphate (6.8 mg/dL) levels. Serum calcium level was 9 mg/dL. Cervical plain radiography showed demineralized lesion affecting the 4th and 5th cervical vertebrae. Magnetic resonance imaging demonstrated bone infiltration of the cervical vertebrae with large paravertebral and intraspinal soft tissue masses with neural compression (Figure 2). Parathyroid

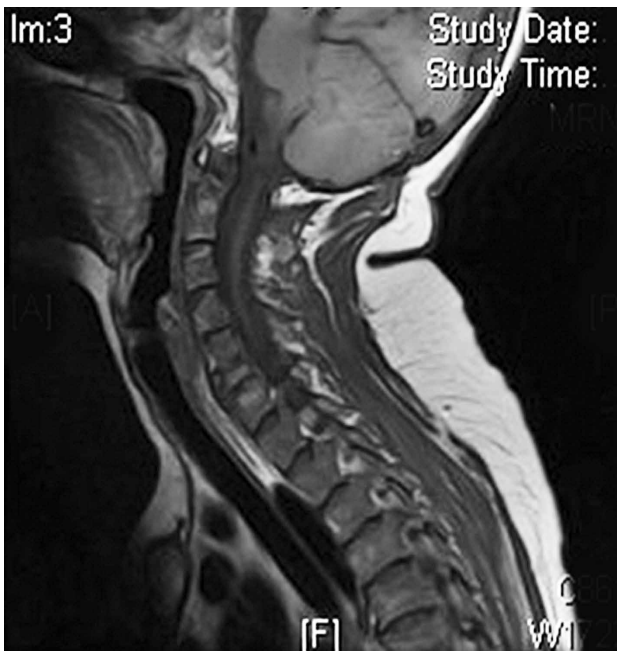

Figure 2. Magnetic resonance imaging demonstrated bone infiltration of the 4th and 5th cervical vertebrae with large paravertebral and intraspinal soft tissue masses and neural compression.

adenomas were documented on ultrasonography.

Surgical removal of the tumor was performed with dural sheet reconstruction after nerve root dissection. Pathologic examination of the excised mass revealed granulation tissue with fibroblasts, histiocytic cells, few plasma cells, and lymphocytes. Numerous osteoclastic giant cells and osteoid-rich bone fragments were present, with microfoci of hemosiderin deposition. Subtotal parathyroidectomy was also carried out. High-dose calcium (both oral and intravenous) was administered for a few months to maintain the serum calcium levels within the normal range. Following surgical intervention, clinical improvement was achieved after 6 weeks with complete mineralization of the spine without nerve defects.

DISCUSSION

Since the first report of brown tumor involving the spine among hemodialysis patients in 1978, another 8 cases have been published.[2-10] The breakdown of these cases revealed that all of them except one were females, with an age range of 24 to 69 years. Concerning the spinal segment, the affected site was thoracic in 6, cervical in 2, and sacral in 1 patient. Regarding the dialysis period, 3 patients had chronic kidney failure who had not started renal replacement therapy yet,[5,7,10] 2 patients suffered from chronic allograft rejection,[3,8] while the remaining 4 patients were on dialysis for 2 to 11 years.

Therapeutic modalities were adopted—in most of the cases—by surgical tumor resection and/or decompression plus parathyroidectomy.[2,5,7,9,10] Nevertheless, steroid therapy was needed in 1 patient[5] and Minerva jacket in another.[8] Another case without neurological features encouraged to be managed by parathyroidectomy preceded by bisphosphonate infusion.[3] This highlights the role of hyperparathyroidism in the pathogenesis of such cases. Thus, parathyroidectomy alone may be enough in the absence of neurological features.[3] The incidence of brown tumor was 1.5% to 13% in patients with kidney failure, according to Griffiths and coworkers (1974)[11] and Sargent and colleagues (1989).[12] Since then and to the best of our knowledge, no reportable incidence was known.

In our report, one of the two patients represented the youngest male reported and even the 1st case with lumbar involvement associated with cauda equina compression, and the 3rd after chronic kidney allograft failure. This patient had many risk factors to develop such a lesion in the lumbar region, such as hyperparathyroidism and possibly previous steroid therapy. The second case represented the 3rd case with symptomatic cervical brown tumor which was managed successfully.

The presentation of brown tumor involvement of spine can be presented either acutely due to cord compression secondary to bone fracture or by progressive features subsequent to the mass effect. The primary goal of treatment is to excise the tumor irrespective of that bone graft is needed or not, together with achieving normal calcium and phosphorus blood levels through medical or surgical treatment of hyperparathyroidism. This plan was attempted in most of the cases including our two patients. The development of brown tumor in the two cases included in our report after a relatively short dialysis period could be attributed to combined effects of steroid therapy received during graft survival and the acute rejection episode and the effect of hyperparathyroidism in addition to unsupported part of the spine (cervical and lumbar), which needed aggressive surgical intervention.

With improvements in the medical care of patients with chronic allograft failure, the number of dialysis-dependent patients, and consequently the incidence of brown tumors, may be expected to increase possibly due to the additive effect of steroid therapy with other comorbidities. Surgical excision might be urgently indicated to save the spine.

CONFLICT OF INTEREST

None declared.

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