

## VERTEBRAL COMPRESSION FRACTURES IN A CHILD: AN UNCOMMON PRESENTATION OF CHILDHOOD ACUTE LEUKEMIA

### BİR ÇOCUKTA VERTEBRA KOMPRESYON KIRIĞI: ÇOCUKLUK ÇAĞI AKUT LÖSEMİNİN NADİR BİR PREZENTASYONU

Ertuğrul KÖSEOĞLU\*, Çağatay ÖZTÜRK\*\*, Ufuk AYDINLI\*\*\*

#### SUMMARY:

We report a case in whom back pain associated with vertebral compressions and collapse were presenting features of childhood acute lymphoblastic leukemia (ALL); radiological remodeling with return of function occurred with remission. This report is intended to promote greater awareness that acute leukemia can cause significant back pain in children without other systemic symptoms.

There should be a high index of suspicion in patients, particularly if there is worsening back pain with the associated radiographic findings of vertebral compression fractures. Antileukemic treatment usually results in rapid symptomatic relief as well as radiographic evidence of bony remodeling.

**Key words:** Vertebra compression fracture, Childhood leukemia, Osteoporosis.

**Level of evidence:** Level IV, Case report

#### ÖZET:

Bu vaka takdiminde, bel ve sırt ağrısı ile birlikte giden vertebral çökme bulgularının akut lenfoblastik lösemi hastalığının öncü bulguları olduğu bir olgu incelendi. Bu yazı ile birlikte, akut lösemnin diğer tipik semptomları olmaksızın sırt ve bel ağrısı ile kendisini gösterebileceği vurgulanmaya çalışıldı.

Sırt ağrısı ve çoklu vertebral çökmeler ile karakterize bir hastada hekim lösemi açısından şüphelenmelidir. Uygun tedavi ile lösemi remisyona girmekte ve radyolojik bulgular normale dönmektedir.

**Anahtar Kelimeler:** Vertebra kompresyon kırığı, Çocukluk çağı lösemisi, Osteoporoz.

**Kanıt Düzeyi:** Düzey IV, olgu sunumu

(\*) Consultant, Uludag University Medical School, Department of Orthopedic Surgery, Bursa.

(\*\*) Consultant, Istanbul Spine Center, Florence Nightingale Hospital, Istanbul.

(\*\*\*) Professor, Uludag University Medical School, Department of Orthopedic Surgery, Bursa.

**Corresponding Address:** Çağatay ÖZTÜRK, MD, Consultant Orthopedic Surgeon Istanbul Spine Center, Florence Nightingale Hospital Abide-I Hürriyet Cad. No: 290, 80220, Şişli, Istanbul, Turkey

**Phone:** +90 (212) 315 36 36

**Fax:** +90 (212) 234 86 89

**e-mail:** cgtyztrk@yahoo.com

## INTRODUCTION:

Acute lymphoblastic leukemia (ALL) is the most common childhood cancer, accounting for about 33 % of all pediatric malignancies<sup>(3)</sup>. ALL is an infiltration of neoplastic cells in the bone marrow, and its peak incidence is around 4 years of age. Clinical presentation usually involves nonspecific symptoms of anorexia, fatigue, or irritability, followed by signs of marrow failure, fever, anemia, bruising, and enlargement of lymph nodes, spleen, and liver<sup>(1,3,7)</sup>. These symptoms are frequently accompanied by musculoskeletal conditions, such as limping, joint pain, and bone pain, meaning these patients often initially present to the orthopedic surgeon. At this initial presentation, many patients have non-specific findings in their laboratory data. Although radiographic changes associated with leukemia in children are often seen in the early stages of this disease, these changes are usually non-specific. Therefore, leukemia with musculoskeletal conditions is frequently misdiagnosed by orthopedic surgeons as juvenile rheumatoid arthritis, septic arthritis, or osteomyelitis, resulting in a delay in the correct diagnosis and appropriate treatment.

Bone and joint pain may be a presenting symptom in around 25 % of patients with acute leukemia<sup>(5-6,15,18,24)</sup>, whereas generalized osteopenia and vertebral complications are less common<sup>(4)</sup>. The literature does not clearly define the incidence of spinal involvement, with only a total of 31 cases located in a review of the literature<sup>(1-2,5-6,13,19)</sup>.

Compression fractures of vertebrae were noted in 20 out of 1.700 children with the acute lymphoblastic leukemia. Usually prognosis in these cases has been favorable (70 % of patients are alive from 5 months to 19 years)<sup>(14)</sup>. Percentage of recovery from compression

fractures has been relatively high. Lymphoblastic leukemia with infiltrations localized in the spine is relatively non-aggressive, develops slowly, and despite extensive lesions to the bones its outcome results are favorable. Main symptoms of spinal involvement include severe and persisting back aches which make walking impossible. These presentations have been associated with slowly evolving, clinically silent leukemia without organomegaly or blasts in the peripheral blood. Such symptoms should indicate the diagnosis of leukemia and advocate proper hematological examinations.

We report a case in whom back pain associated with vertebral compressions and collapse were presenting features of childhood acute lymphoblastic leukemia (ALL); radiological remodeling with return of function occurred with remission. This report is intended to promote greater awareness that acute leukemia can cause significant back pain in children without other systemic symptoms.

## CASE REPORT:

A 6-year-old girl was admitted with a one month history of progressive back pain following a minor trauma. On initial examination, she appeared to be in pain, lying on her side. Vital signs were a heart rate 92 beats per minute, blood pressure of 115/65 mm Hg, respiratory rate (RR) 24/min, and a temperature of 37.1° C taken orally. The head and neck examinations showed no significant adenopathy or mucosal abnormalities. No organomegaly or abnormal axillary and inguinal lymph nodes were found. Heart sounds were normal, and lungs were clear on auscultation. The spine examination was limited because of pain, but it revealed

tenderness on palpation of the spinous processes of the dorsal and lumbar vertebrae. Passive movements of both legs and arms were normal. No neurological abnormalities were found on initial or subsequent examinations.

Conventional x-ray of the spine showed mild compression on several thoracic and lumbar vertebrae (Figure-1) and extended vertebral demineralization. Whole spine magnetic resonance imaging (MRI) showed thoracic 7-8-9-12 and lumbar 1-2-3-4 vertebral collapse (Figure-2). There was diffuse signal reduction and hypointense appearance of vertebral bodies on T1-weighted and T2-weighted sequences; suggesting lymphoproliferative disorders. Bone mineral density revealed osteoporosis at lumbar vertebrae (total T score was -5.9).



**Figure-1.** Conventional lateral x-ray of the spine showed mild compression on several thoracic and lumbar vertebrae.

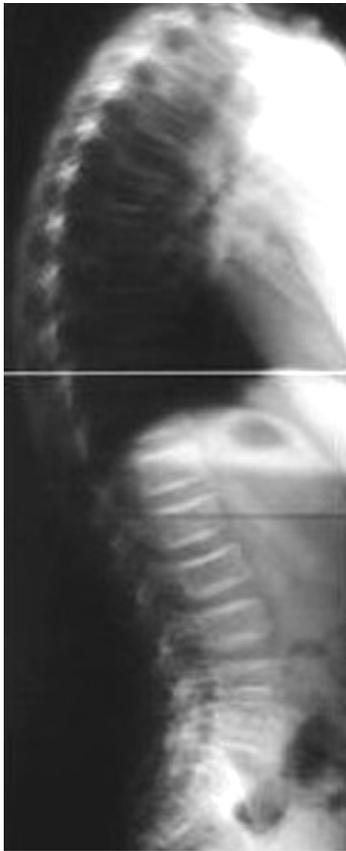


**Figure-2.** Sagittal whole spine magnetic resonance imaging (MRI) showed thoracic 7-8-9-12 and lumbar 1-2-3-4 vertebral collapse.

Initial peripheral blood count showed hemoglobin value of 8.3 g/dl (normal value: 12.0-18.0 g/dl), white blood cell count of 1.46 K/ $\mu$ L (normal value: 5.2-12.4 K/ $\mu$ L) and platelet count of 229 K/ $\mu$ L (normal value: 150-300 K/ $\mu$ L). On blood chemistry, calcium concentration was 9.3 mg/dl (normal value: 8.5-10.5 mg/dl), phosphate 5.3 mg/dl (normal value: 2.0-5.0 mg/dl) were detected. Abdominal ultrasonography revealed no abnormal findings.

Following bone marrow aspiration, diagnosis was uncertain; therefore bone marrow biopsy was performed and diagnosis of acute lymphoblastic leukemia (ALL) was confirmed.

Induction treatment with prednisone, vincristine and daunorubicin was started. The girl improved rapidly and the pain resolved. A thoracolumbosacral orthosis was used for 3 months and the activities were restricted. Eighteen months after first presentation, there was no back pain and spontaneous remodeling of vertebrae had occurred with restoration of vertebral height and shape without any kyphotic deformity (Figure-3). She continues to remain pain free and in remission on treatment.



**Figure-3.** 18 months after first presentation, there was no back pain and spontaneous remodeling of vertebrae had occurred with restoration of vertebral height and shape without any kyphotic deformity.

#### DISCUSSION:

Leukemia is the most common form of cancer in children. The onset of leukemia is often

insidious. Bone involvement is a frequent phenomenon in children with leukemia and bone pain is caused by massive proliferation of hematopoietic tissue within medullary cavities, most commonly in long bones and vertebral bodies. The presence of bony involvement is associated with a favorable prognosis <sup>(12)</sup>. The development of osteoporosis, predominantly central with collapse or compression of several vertebral bodies, however, is a rare occurrence, seen in less than 1% of children <sup>(1-2,5-6,9-11,15-16,19-20)</sup>.

This extensive skeletal involvement can develop before leukemia becomes overt and may be associated with delayed diagnosis <sup>(19)</sup>. Characteristically, there is no organomegaly or adenopathy, no blasts in the peripheral blood or spinal fluid, and a normal chest radiograph <sup>(2)</sup>; blood findings include moderate anemia and a low white cell count with lymphocytosis. Bone metabolic parameters are usually normal, but hypercalcemia may be seen <sup>(8)</sup>.

Pain is most commonly reported in the lower thoracic and upper lumbar region. Observations have shown that vertebral fractures are often seen in cases of ALL with hypercalcemia and low blast cell counts, which were not noted in our patient <sup>(19)</sup>. Vertebral compression fractures seldom cause neurological compromise, and they appear to remodel as the underlying disease is treated. Leukemic remission normally correlates with the disappearance of pain and a return to normal function. Chemotherapeutic agents and osteoporosis secondary to disuse may account for fractures seen during the treatment.

MRI is highly sensitive to changes in bone marrow composition and it plays a role as a modality to confirm hematological malignancies <sup>(21)</sup>. Furthermore its role in monitoring response

to treatment as well as in diagnosing complications is well established<sup>(17,22-23)</sup>. A decrease in signal at T1-weighted images lower than the disc or muscles is the most pronounced feature and should be considered abnormal.

There should be a high index of suspicion in patients, particularly if there is worsening back pain with the associated radiographic findings of vertebral compression fractures. Antileukemic treatment usually results in rapid symptomatic relief as well as radiographic evidence of bony remodeling.

#### REFERENCES:

1. Bjerregaard LL, Rosthoj S. Vertebral compression and eosinophilia in a child with acute lymphatic leukemia. *J Pediatr Hematology/Oncology* 2002; 24 (4): 313-315.
2. Blatt J, Martini SL, Panchansky L. Characteristics of lymphoblastic leukemia in children with osteopenia and vertebral compression fractures. *J Pediatr* 1984; 105: 280-282.
3. Carriere C, McManus BC. Vertebral fractures as initial signs for acute lymphoblastic leukemia. *Pediatr Emergency Care* 2001; 17 (4): 258-261.
4. Cohn SL, Morgan ER, Mallette LE. The spectrum of metabolic bone disease in lymphoblastic leukemia. *Cancer* 1987; 59: 346-350.
5. Epstein BS. Vertebral changes in childhood leukemia. *Radiology* 1957; 68: 65-69.
6. Kayser R, Mahlfeld K, Nebelung W, et al. Vertebral collapse and normal peripheral blood cell count at the onset of acute lymphatic leukemia in childhood. *J Pediatr Orthop B* 2000; 9 (1): 55-57.
7. Kobayashia D, Satsumaa S, Kamegayab M, et al. Musculoskeletal conditions of acute leukemia and malignant lymphoma in children. *J Pediatr Orthop B* 2005; 14: 156-161.
8. Leheup B, Membre H, Gerard H, et al. Lymphoblastic leukemia with osteopenia and vertebral compression fractures. *J Pediatr* 1985; 106: 160.
9. Mandel R, Vic P, Nelken B, et al. Vertebral compression revealing acute lymphoblastic leukemia. *Arch Pediatr* 1996; 3 (5): 466-469.
10. Meehan PL, Viroslav S, Schmitt EW Jr. Vertebral collapse in childhood leukemia. *J Pediatr Orthop* 1995; 15 (5): 592-595.
11. Mehrotra S, Kumar A, Singh BN. Vertebral compression fracture. An unusual presentation of childhood acute lymphoblastic leukaemia. *J Assoc Physicians India* 1990; 38 (12): 943-945.
12. Müller HL, Horwitz AE, Kühl J. Acute lymphatic leukemia with severe skeletal involvement. A subset of childhood leukemia with a good prognosis. *Pediatr Hematol Oncol* 1998; 15: 121-133.
13. Newmann AJ, Melhorn DK. Vertebral compression in childhood leukemia. *Am J Dis Child* 1973; 125: 863-865.
14. Ochocka M, Matysiak M, Armata J, et al. Compression fractures of the vertebrae in children with acute lymphoblastic leukemia. *Pol Tyg Lek* 1992; 47 (16-17): 54-56.
15. Pandya NA, Meller ST, MacVicar D, et al. Vertebral compression fractures in acute lymphoblastic leukaemia and remodelling after treatment. *Arch Dis Child* 2001; 85: 492-493.
16. Parker BR, Marglin S, Castellino RA. Skeletal manifestations of leukaemia. Hodgkin's disease and non-Hodgkin's lymphoma. *Semin Roentgenol* 1980; 15: 302-315.

17. Pieters R, van Brenk AI, Veerman AJ. Bone marrow magnetic resonance studies in childhood leukemia. Evaluation of osteonecrosis. *Cancer* 1987; 60: 2994-3000.
18. Rogalsky RJ, Black GB, Reed MH. Orthopedic manifestations of leukemia in children. *J. Bone Joint Surg. Am.* 1986; 68 (4): 494-501.
19. Samuda GM, Cheng MY, Yeung CY. Back pain and vertebral compression: an uncommon presentation of childhood acute lymphoblastic leukemia. *J Pediatr Orthop* 1987; 7 (2):175-178.
20. Santangelo JR, Thomson JD. Childhood leukemia presenting with back pain and vertebral compression fractures. *Am J Orthop* 1999; 28 (4): 257-260.
21. Takagi S, Tanaka O. The role of magnetic resonance imaging in the diagnosis and monitoring of myelodysplastic syndromes or leukemia. *Leuk Lymphoma* 1996; 23: 443-450.
22. Tardivon AA, Vanel D, Munck JN, et al. Magnetic resonance imaging of the bone marrow in lymphomas and leukemias. *Leuk Lymphoma* 1997; 25: 55-68.
23. Van Zanten TE, Golding RP, Taets-van Amerongen AH, et al. Nuclear magnetic resonance imaging of bone marrow in childhood leukaemia. *Clin Radiol* 1988; 39: 77-81.
24. Vassilipoulou-Selin R, Ramirez I. Severe osteopenia and vertebral compression fractures after complete remission in an adolescent with acute leukaemia. *Am J Hematol* 1992; 39: 142-143.