Intractable Neck Pain Due to Osteoblastoma: A Case Report and Review of the Literature

Osteoblastomaya Bağlı Kontrol Altına Alınamayan Boyun Ağrısı: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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Summary

A 12 year-old girl presented with a 10-month-history of the left lateral neck pain. The patient had been examined by eight physicians and multiple diagnoses had been made before. Despite treatments, neck pain had not been relieved and the intensity of the pain had gradually increased. On admission, there was local tenderness on the left side of the neck and cervical movements were restricted. The neurological examination and routine laboratory tests were normal. Except for decreasing cervical lordosis, of previous plain radiographs and magnetic resonance images of the cervical spine were normal. Based on clinical signs and symptoms, we suspected that the patient had a cervical mass. A soft tissue ultrasonography of the neck showed a suspicious bony hypertrophy or calcification-like lesion in the level of the fourth cervical vertebra. The subsequent computed tomography scanning confirmed a tumour in the left transverse process of the fourth cervical vertebra. After the tumour was excised, its pathological examination revealed osteoblastoma. Neck pain disappeared after surgery. Recurrence was not seen in the final follow-up. Careful clinical and radiologic evaluation should be performed in all pediatric patients with neck pain to rule out bone tumours. Although osteoblastoma is rare, diagnosis is commonly delayed and effective treatment is important to prevent neurologic complications and recurrence. *Turk J Phys Med Rehab 2009;55:89-93.*
Key Words: Neck pain, cervical spine tumours and osteoblastoma

Özet

On iki yaşında kız çocuğu, 10 aydır boyun sol tarafında ağrı şikayeti ile başvurdu. Hasta daha once 8 hekim tarafından değerlendirilmiş ve çok sayıda tanı almıştı. Önceki tedavilere rağmen, boyun ağrısı geçmemiş ve ağrı yoğunluğu giderek artmıştı. Bize müracaat ettiğinde, boyun sol tarafında lokal hassasiyeti vardı ve boyun hareketleri kısıtlıydı. Nörolojik muayene ve rutin laboratuvar incelemeleri normaldi. Lordozda azalma hariç servikal omurganın daha önceki direk grafileri ve manyetik rezonans görüntüleri normaldi. Klinik semptom ve bulgulara dayanarak, hastada servikal kitle olduğundan şüphelendik. Boyun yumuşak doku ultrasonografisi 4. servikal vertebra seviyesinde şüpheli kemik hipertrofisi veya kalsifikasyon benzeri bir lezyon olduğunu gösterdi. Daha sonraki bilgisayarlı tomografi incelemesi 4. servikal vertebranın sol transvers çıkıntısında tümür varlığını doğruladı. Tümör çıkarıldıktan sonraki patolojik incelemesi osteoblastoma olduğunu gösterdi. Cerrahi sonrası hastanın boyun ağrısı geçti. Son takipte rekürrens görülmedi. Boyun ağrılı tüm çocuk hastalarda kemik tümörlerini dışlamak için dikkatli klinik ve radyolojik değerlendirme yapılmalıdır. Osteoblastoma nadir olmasına rağmen, tanıda gecikme yaygındır, rekürrensi ve nörolojik komplikasyonları önlemek için etkili tedavi önemlidir. *Türk Fiz Tıp Rehab Derg 2009;55:89-93.*
Anahtar Kelimeler: Boyun ağrısı, servikal omurga tümörleri ve osteoblastoma

Introduction

Benign osteoblastoma was independently described for the first time by Jaffe and Lichfenstein in 1956 as a vascular, osteoid and bone-forming tumour cytologically characterized by osteoblasts (1-2).

Osteoblastoma accounts for less than 1% of all primary bone tumours and approximately 10% of all benign spinal tumours (3). The tumour is most often located in the posterior elements of the vertebrae (4). Males younger than 20 years are affected more often than females (3). The most frequent clinical feature of osteoblastoma is pain, followed by torticollis,

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scoliosis, and neurologic symptoms (5,6). The tumour invades into the epidural space and nerve roots and causes compression on the cord in most of the cases (7).

The treatment of choice for osteoblastoma is complete surgical resection. The tumour has been reported to recur in up to 10% of the cases, especially in case of inadequate primary excision (8,9).

We report a case of intractable neck pain due to osteoblastoma involving the cervical vertebrae. It is interesting that it is rare and with lacked specific symptoms and signs and therefore had been overlooked before and that the outcome of surgical intervention was excellent.

Case Report

A 12 year-old girl presented with a 10-month-history of left lateral neck pain, without radiation to the shoulder, back and arm. She also denied concomitant headache, blurred vision, weakness, numbness, tingling or bowel and bladder dysfunction. There was no associated history of weight loss, fever, night sweats, diaphoresis or prior trauma.

The patient had been examined by eight physicians from various specializations and multiple diagnoses had been made including "sternocleidomastoid muscle strain", "myofascial syndrome", "cervical radiculopathy", "joint subluxation", "occipital neuralgia" and "somatoform disorder" before referring to our clinic. She had received nonsteroidal anti-inflammatory drugs, myorelaxants, antidepressants (sertralin, amitriptyline) and anxiolytics (diazepam, alprazolam). Despite symptomatic treatment, neck pain was not relieved and the intensity of the pain gradually increased. In addition, the patient had received 15 sessions of physical therapy employing heat, TENS, neck strengthening and flexibility exercises, but the treatment had failed.

Physical examination showed no cervical deformity such as scoliosis or torticollis. Tenderness was localized in the left side of the neck under the mid one-third of the sternocleidomastoid muscle. Although the localized pain was significantly increased by direct palpation, a mass lesion could not be identified. Passive cervical spinal movements were painful and restricted in all directions. Especially neck pain was increased by cervical rotation to the left. Spurling tests provoked no pain on both sides. Compression and distraction tests were positive. The neurologic examination was also normal, with intact cranial nerves, 5/5 motor strength in all limbs, normoactive symmetric muscle stretch reflexes, normal sensory and coordination testing. All pathologic reflexes were negative bilaterally. There was no evidence of carotid or subclavian artery bruits.

Routine laboratory examinations showed no abnormality. The plain radiographs of the cervical spine were normal except for decreasing cervical lordosis (Figure 1a/b). In addition, previous magnetic resonance images (MRI) of the cervical vertebrae (Figure 2a/b) and the brain, cerebral MR angiography and electroencephalography were normal. Based on clinical signs and symptoms, we suspected that the patient had a cervical mass. Soft tissue ultrasonography (USG) of the left cervical region indicated suspicious bony hypertrophy or a focal, calcification-like lesion at the level of the fourth cervical vertebrae. The subsequent computed tomography (CT) examination of the cervical spine showed a heterogeneously sclerotic, nodular, bony lesion, 1.5x1.3 cm in size. (Figure 3). The lesion seemed to originate from the left transverse process of the fourth cervical vertebrae, partially extending to the

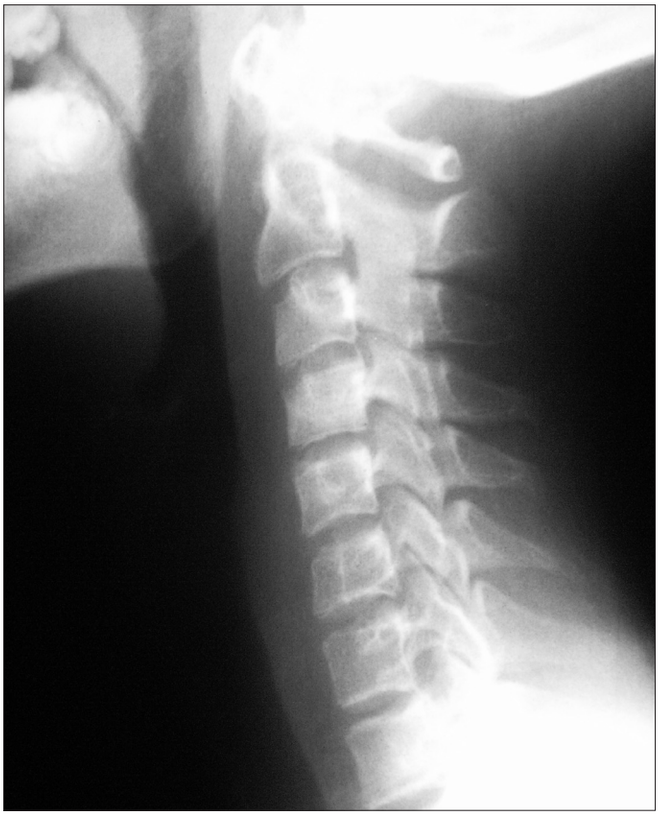

Figure 1a. Lateral plain graphy of the cervical spine; decreased lordosis.

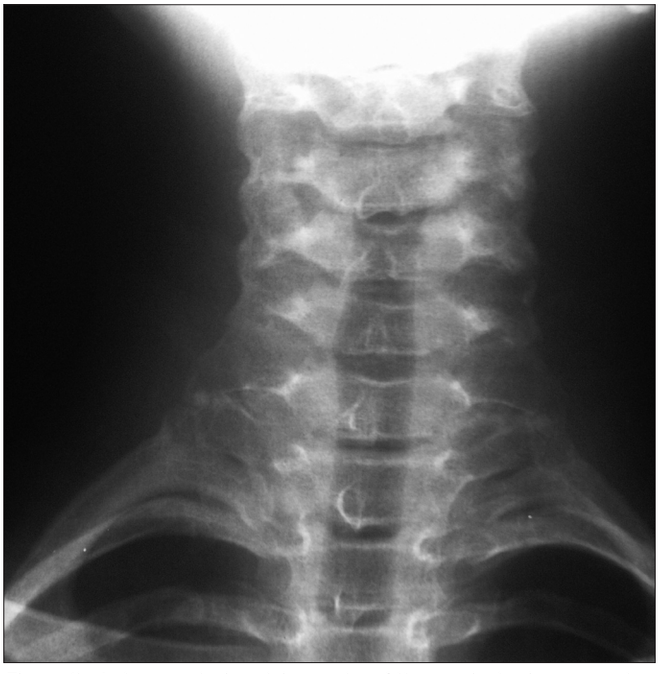

Figure 1b. Antero-posterior plain graphy of the cervical spine; normal.

vertebral corpus and narrowing the left transverse and intervertebral foramen without compression and displacement of the spinal cord. Osteoid osteoma, osteochondroma or osteoblastoma was diagnosed radiologically. The patient was referred to a spinal surgeon. Vertebral artery angiography performed before surgery showed no sign of compression on the left vertebral artery.

On operation, an anterior approach was used for the spine and the left foramina and the corpus of the fourth cervical vertebra was exposed. Then, the tumour mass was totally excised. Pathological examination of the surgical specimen revealed osteoblastoma (Figure 4).

Neck pain was significantly relieved after surgery. Twelve months after surgery, radiologic or clinic signs of local recurrence were not identified.

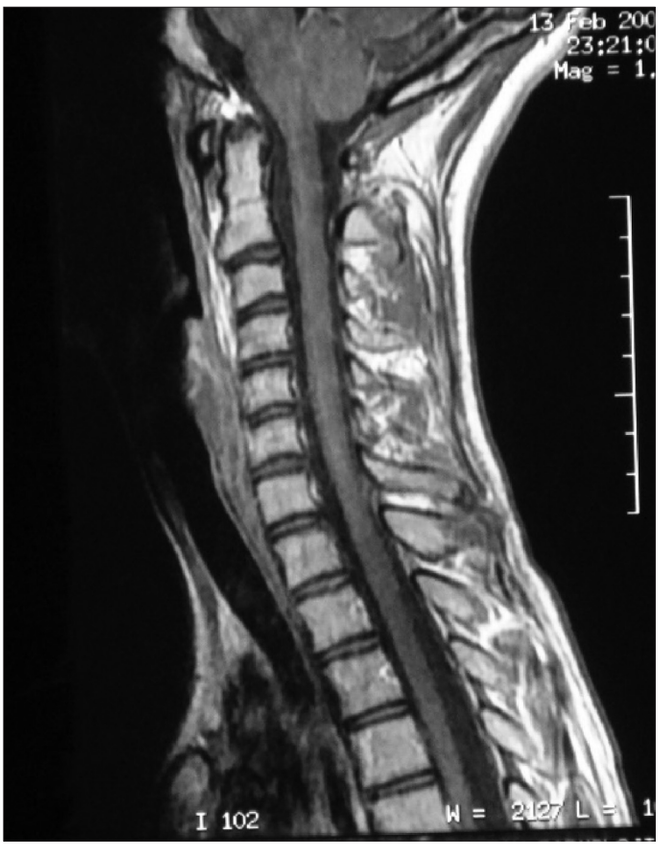

Figure 2a. Magnetic resonance image of the cervical spine.

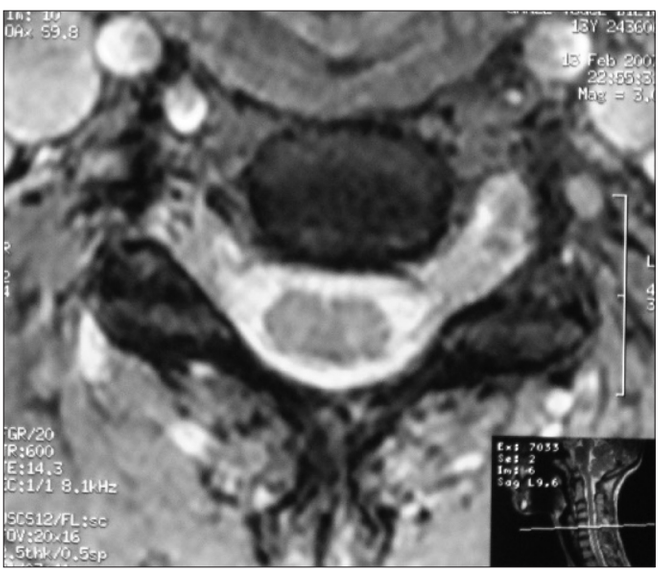

Figure 2b. Magnetic resonance image of the cervical spine.

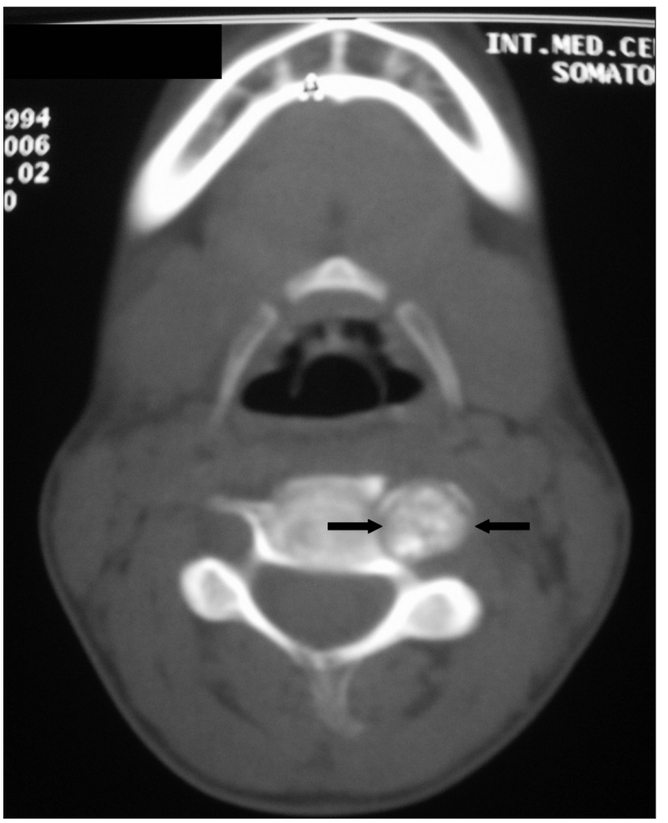

Figure 3. CT image of the cervical vertebrae; a heterogeneously sclerotic, nodular bony lesion in the transverse process of the fourth cervical vertebrae (arrrows).

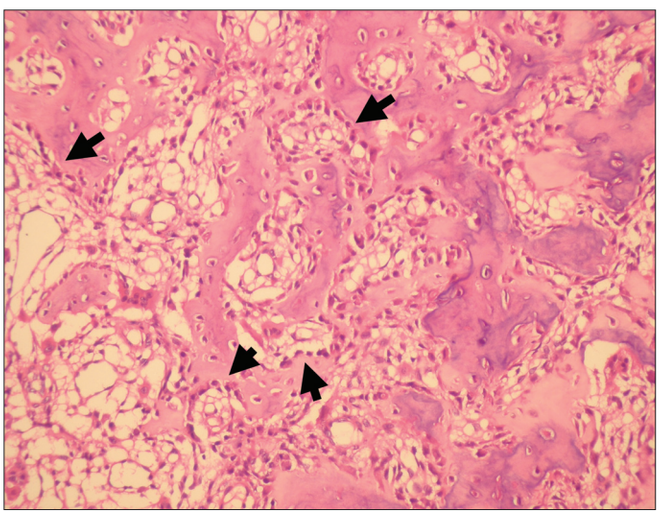

Figure 4. A view of bone lamellae surrounded by active osteoblasts (arrows) (H&E, X200).

Discussion

Osteoblastoma is a rare benign tumour. Spinal involvement is described in 30% to 40% of all cases (3,5). In 20% to 40% of the patients with spinal involvement, the cervical spine is affected (6,9).

As in the case presented here, when located in the spine, the tumour frequently occurs in the lamina, pedicles, or in the transverse and spinous processes (3-5). Although the case presented here is a 12 year-old girl, males are affected more often than females. In fact, it is striking that the male-to-female ratio is about 2.5 to 1. It occurs predominantly in adolescents and adults in their twenties (3).

As in all skeletal neoplasms, dull, local, gradually increasing pain and stiffness are usually the main clinical symptoms (3,10). In a series of Bruneau et al. (11) all of the seven patients complained of pain and three patients presented with neck stiffness. Kirwan et al. (12) reported that all patients had pain, and 16 of 18 patients (89%) had spinal stiffness. Consistent with the literature, the case reported here also presented with neck pain and stiffness.

Painful scoliosis and torticollis are well recognized signs of spinal osteoblastoma (13). Raskas et al. (6) reported that approximately 45% of the patients with cervical lesions initially had torticollis. In a series of 29 patients with cervical osteoblastoma Nemoto et al. (3) showed that 17% of the patients had torticollis. Scoliosis is induced by muscular spasm secondary to an inflammatory effect around the tumour (14). Kirwan et al. (12) showed that all patients had scoliosis at the time of presentation. Some authors presented that 25% to 62% of patients had scoliosis (3,7). In addition, Saifuddin and Ozaki et al. (15,16) reported that 63% and 77% of the patients had scoliosis respectively.

Scoliosis, in the long term, may not be relieved with treatment because of damage to the growth plate and degeneration or fibrosis of the muscle by chronic inflammation (16). Pettine et al. (7) reported that most patients with a short duration (less than 15 months) of symptoms had improvement of scoliosis after surgery. However, patients with over 15 months of symptoms did not have improvement.

Neurologic deficits such as muscle weakness, sensory disturbance, changes in reflexes and even neurogenic bladder, tetraparesis, and paraparesis are often seen in patients with osteoblastoma (13,16). Boriani et al. (5) showed that 75% of the patients with thoracic lesions (6 of 8 patients) had paraparesis. Similarly, Ozaki and Zileli et al. (16,17) reported that 69% of the patients had neurologic symptoms. In the present case, there was no cervical deformity such as scoliosis and torticollis. There was only local tenderness on the neck and restricted cervical movements. In addition, the neurologic examination was also normal. Delay in the diagnosis is very common, and seems to occur in six-twelve months on average. However, Kirwan et al. (12) presented that the average delay was 19 months. In our case, the nonspecificity of the symptoms and signs, which had been neglected by many physicians, contributed to the delay in diagnosis.

Diagnostic imaging routinely includes plain radiographs, three phase bone scan, CT and MRI (18). Osteoblastoma of the spine may usually be detected by a high-quality plain radiograph. Classical plain radiographic investigation shows radiolucent destructions with a perifocal sclerosis. As radiologic abnormalities are often late in appearance, a definitive diagnosis may be delayed for more than one year (3,19-21). In agreement with the literature, the tumour could not be detected on plain radiographs in the case presented here. If plain radiographs are not sufficient to diagnose an osteoblastoma, the CT scans and MRI are of great value in diagnosing the tumor and detecting the location and the extent of both soft tissue and osseous involvement (22). Compared with MRI, CT clearly shows cortical destruction and identifies matrix mineralization. Therefore, Ozkal and Shaikh et al. (22,23) suggested that CT is superior to MRI in terms of both characterization of the tumour and accuracy of local staging.

In our case the tumour could not be demonstrated by MRI, but CT clearly showed the tumour and its exact origin and extent. Previously, LaBan and Riutta (24) reported a similar case of osteoblastoma presenting with neck pain. In their case, although plain radiographs and MRI of the cervical spine were normal, CT and bone scanning showed an osteoblastoma.

Before CT scanning, we performed soft tissue USG and suspected a lesion on the fourth cervical vertebra. Although previous studies have shown that sonographically guided core needle biopsy is very reliable and accurate for most bone tumours with extraosseous masses located in appendicular skeleton, we could find no evidence regarding the role of soft tissue USG in the diagnosis of osteoblastoma (25,26). Three-phase bone scanning shows increased tracer uptake at the site of the lesion (18). A large tumour can be associated with compression of the vertebral arteries. Zambelli et al. (27) recommended that the vertebral arteries should be preserved in all cases. Angiography in the current case showed no sign of compression.

Cervical osteoblastoma are treated for persistent pain, increase in size or the presence of a neurologic deficit. The treatment of choice for osteoblastoma of the spine is complete surgical resection (4,28). It has been reported to recur in up to 10% of the cases due to partial resection of the tumour (7,9,10). For recurrent osteoblastoma, the mainstay of therapy is re-excision (16). If complete resection is not possible, radiotherapy and in some cases chemotherapy seem to be alternative treatment options (28,29) In the current case, symptoms completely disappeared and there were no recurrent manifestations of the tumour on MRI twelve months after surgery.

In conclusion, careful clinical and radiologic evaluation should be performed in all pediatric patients with neck pain lasting for extended periods of time to rule out bone tumours. Although osteoblastoma is a rare tumour, delay in its diagnosis is common and effective treatment is important to prevent neurologic complications and recurrence.

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