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OLGU SUNUMU / *CASE REPORT*

CERVICAL SOLITARY OSTEOCHONDROMA A CASE REPORT

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Abstract:

Solitary osteochondroma, despite being the most frequent benign lesion of the skeletal system, rarely affects the cervical spine. In this paper, we report on a 12-year-old boy admitted with a mass at his neck, with a cecile exostoses at the 4th cervical vertebrae. Radiographic, MRI and CT scan revealed anterior wedging and spontaneous fusion at the 4th and 5th cervical vertebrae. Surgical excision of the mass was performed and posterior fusion was added to prevent potential cervical kyphosis. Postoperatively, a cervical collar was used for two months and a solid fusion mass was noted at the postoperative 6th month. The surgical treatment and natural features of this rare disease are discussed in this case report.

Key words: *Osteochondroma, cervical tumors, multiple exostoses, and surgical treatment.*

Özet:

İskelet sisteminin en sık görülen iyi huylu lezyonu olmasına karşın soliter osteokondromlar servikal omurgada nadiren yerleşim gösterir. Bu yazıda boynunda kitle ile başvuran ve dördüncü servikal omurgasında sesil osteokondrom saptanan 12 yaşında bir erkek çocuk sunmaktayız. Radyografi, magnetik rezonans ve bilgisayarlı tomografi incelemelerinde C4 ve C5 omurgalarda anterior kamalaşma ve spontan füzyon saptandı, Kitle eksize edildi ve potansiyel cerrahi sonrası kifozu önlemek için posterior füzyon eklendi. Postoperatif dönemde iki ay boyunluk kullanıldı ve altıncı ayda füzyon sağlandı. Bu olgu sunumunda soliter osteokondormun bu nadir yerleşiminin bulguları ve cerrahi tedavisi tartışılmaktadır.

Anahtar kelimeler: *Osteokondroma, servikal tümör, multipl egzostozis, cerrahi tedavi.*

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INTRODUCTION

Osteochondromas are the most common benign tumor of bone and they account for approximately one-third of the skeletal benign tumors[14] They can be broad based or have a pedicle that separates them from bone. Macroscopically, they have a cartilaginous cap. They are usually found on the metaphysis of long bones such as distal femur, proximal tibia and proximal humerus. They can be also observed on flat bones such as scapula, ilium and costae. Spine is rarely affected. In the cervical spinal involvement serious neurological symptoms can appear because of the mass effect on the neurovascular structures and soft tissues [8. 16. 33].

The most common localization is posterior colon in the cervical spinal lesion [14]. It has been reported that in the cases in which the tumor is on the lamina of the cervical vertebrae and has neurological deficit because of the spinal cord invasion. Wide decompression can cause vertebral instability and kyphosis can occur after postlaminectomy. In these situations, it has been reported that surgical methods such as posterior fusion, anterior and posterior instrumentation can be useful in preventing these complications [3.10. 12]. In this case report, we present a twelve-year-old patient admitted with a mass at his neck as this involvement is uncommon and as far as we know there is no report on anterior wedging and spontaneous anterior fusion at the same level in the literature.

CASE REPORT

A twelve years old fatty boy was admitted our hospital with a mass on his back of neck. A doctor discovered the palpable mass and referred the patient to us. In physical examination, there was no pathological finding except a palpable solid lesion. In radiological examination, there was irregularity in spinous processes of 4th and 5th cervical vertebrae (Figure-1.a-b).

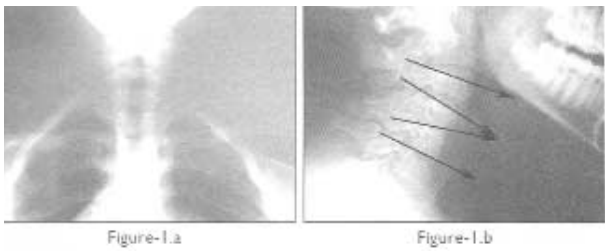

Figure-1.a-b. In the preoperative cervical anterior-posterior and lateral grafics of the patient, solitary osteochondroma originating from the spinous process of C4 vertebra and anterior wedging and spontaneous anterior fusion are seen between C4 and C5 vertebrae.

In neurological examination, cranial nerves were intact and there was no motor or sensorial deficit. Cervical computerized tomography (CT) showed that the lesion was pedinculed exostoses of the spinous process of 4th cervical vertebra.

We observed a lesion which is at C4 vertebre and has reached subcutaneous fat tissue near the C5 spinous process from the paravertebral muscle tissue in the cervical magnetic resonance imagination (MRI). Besides, there was no contrast enhancement and its dimensions were 25x 15x 15 mm. The anterior intervertebral disc spaces were decreased at C4-5 vertebrae and there was anterior fusion between these vertebrae (Figure-1. c-d).

Due to these findings, cervical plain lateral radiography was taken once more. The intersegmental angle between C4-5 vertebrae was increased to the degree of kyphosis and reached 18° but didn't destroy the cervical sagittal lordotic contour.

In the detailed skeletal system examination, there was a painless mass on the back of the patient's neck, and the range of motion was full. There was no exostoses in other parts of his body. There were no characteristic findings in his family story, either. According to these findings and symptoms, the patient was diagnosed with solitary cervical osteochondroma. We plan-

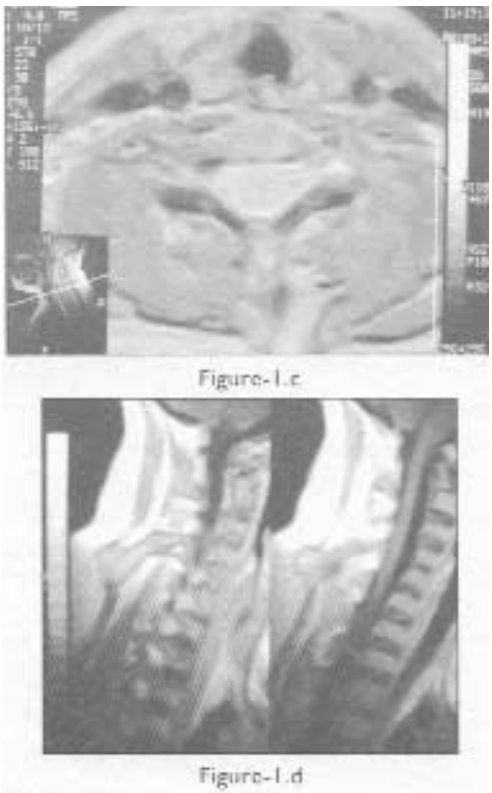

Figure-1.c-d: Preoperative sagittal and axial MRI sections.

ned surgical excision on account of the posteriorly localized lesion that caused anterior wedging and fusion by mechanical effect.

With the patient placed in prone position, we performed posterior midline incision. After dissection of C3-6 posterior structures, the lesion was totally excised with a part of C4 spinous process. After decortications of laminas of C4-5, short segment posterior fusion with autologeos iliac spongious graft was performed (Figure-1.e, f).

Histopathological examination semonstrated abone tissue that had a pedicle and regular gray blue cartilaginous cap. Microscopically, there was trabecular bone structure, which had active enchondral ossification and thin pink fibrin capsular surface and mature cartilaginous areas. In view of these findings, the diagnosis was definitely solitary osteochondroma (Figure-2.a, b)

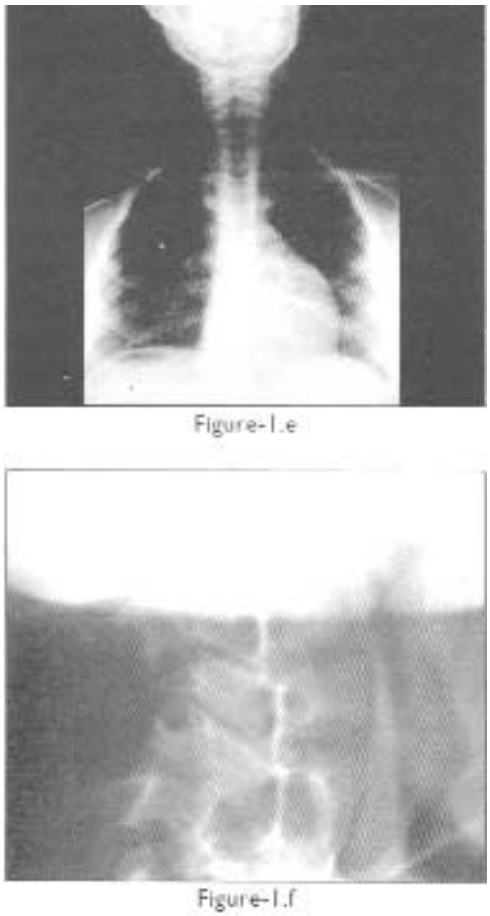

Figure-1.e-f: In the early postoperative radiographs of the patient, excission of the tumor mass can be seen.

Postoperatively, a cervical collar was used for two months and after then, we allowed free neck motions. A solid fusion mass was noted at the postoperative 6th month and there was no recurrence (Figure-1. g-h).

DISCUSSION

Osteochondromas account for 10 % of bone tumors and 36-41 % of benign bone tumors [16.25]. This term was first used in Liechtenstein classification. Because of enchondral ossification, it is thought that this tumor originates from periostal tissue. Today, most of the pathologists think that this tumor is a hamartomatous lesion, which occurs with aberrant growing in the growing plates[16].

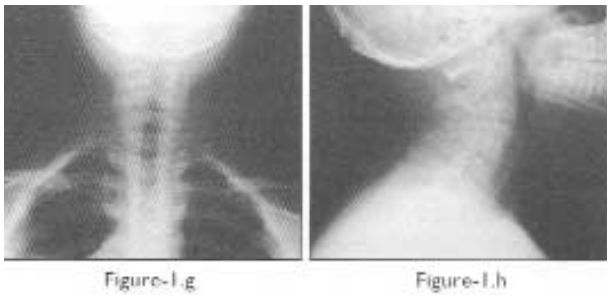

Figure-1.g-h: In the cervical radiograph at postoperative 6th month (g, h) solid fusion mass can be observed.

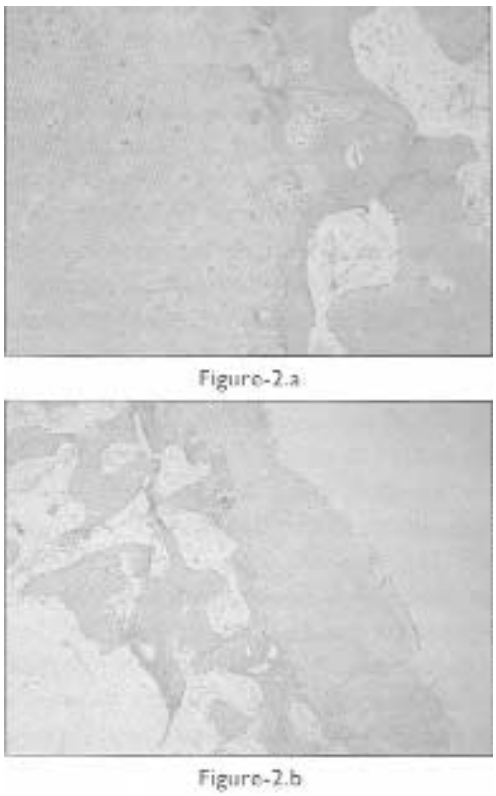

Figure-2. a-b: Gross appearance of the material obtained with excisional biopsy (a). In histopathological examination, active endochondral ossification areas seen are surrounded by cartilage cell areas, covered by fibrous membrane (H. E., x 100),

Although skeletal system is a common localization, vertebral involvement is about 3-7 %. Only 1-3 % of cases have cervical vertebral involvement [15]. The tumor usually occurs at cervical or upper thoracic area in the symptomatic patients as in our case[14]. Gottlieb et al. reported that 91 % of the spinal osteochondromas are at upper level of the 5th thoracic vertebrae[17]. According to Albrecht et al. 34 % of these cases are found at lumbar vertebrae, and 7 % at sacral vertebrae[2]. Roblot et al. report a patient with thoracic involvement[31].

More than 2000 cases, who had multiple exostoses with autosomal dominant inheritance so far, have been reported[11]. Cervical involvement occurred in 7-9 % of these cases [18]. Albrecht et al. state that 130 cases were reported in the literature between 1907 and 1992 with multiple solitary (one bone, one lesion) involvement and 1-4 % of them were at vertebrae. Average age of patients was 21.6 in multiple lesions and 30 in solitary lesions. In all of these groups the vertebral localization was in cervical area in more than one half of the patients and the male/female ratio was 1/ 2.5 [l 6.25]. In this recent study, we have reported a 12 years old male patient who had only one solitary cervical lesion and there was no history of such a case in his family.

Osteochondromas may develop on vertebral body, neural arch and transverse process. They can be stalked or broad based on the bone surfaces [14]. Most of the lesions occur on posterior colon with cervical involvement according to Knoeller et al[22]. Nielsen et al. described a case in whom the lesion was localized on corpus of 2nd cervical vertebrae and Trainee et al. described another case with the lesion localized on the spinous process of 6th cervical vertebrae [28.36]. In our recent study, the case had pedinculed lesion, which was on spinous process of 4th cervical vertebrae. It is pointed out that it is seldom seen.

Many of the osteochondromas are asymptomatic and are discovered incidentally. The most frequent symptoms are pain, neurovascular

dysfunction, limitation of motion and palpable mass [16,32]. Sudden death due to odontoid involvement has been reported [7]. Pain and dysfunction occur because of the mass effect of the tumor. Frequently pain develops because of the bursa formation on the lesion, neurovascular impingement, limitation of the range of motion and malign transformation. It is suggested that malignant transformation should be eliminated if pain appears [14,25]. According to Gitelis and Sorepanth 27 % of the cases had bursa formation, 23 % limitation of motion, 10 % neurological symptoms and 7 % had vascular injury [16]. Limitation of neck motion can occur in C1-C2 vertebral involvement. Cherubino et al. reported a 37 years old case with C1-C2 involvement and limitation of neck motion disappeared after the excision of the lesion [6].

Reid originally described the neurovascular involvement in 1843. In 1907, Oschener performed laminectomy to a 3-year-old patient with C2 involvement [16]. The cases with neurological symptoms are rare, because compression on neurological structures is rarely seen in vertebral involvement. But spinal cord compression and myelomalasia was found in 47 % of reported symptomatic cases[14]. Govender and Parbhoo reported that the most common symptoms were neurological dysfunction and myelopathy in 117 symptomatic patients who had solitary or multiple hereditary osteochondromas between 1943 and 1997 [18]. Albrecht et al reported that the cord compression occurred twice as much in hereditary types as in solitary types [2]. Khosla et al. mentioned that 41 solitary osteochondromas with neurological findings were reported until 1999[21]. Osteochondromas grow parallel to skeletal growth and when the spinal canal diameter growth stops, cord impression can occur[14]. Depending on the level of cord impression, the cases with tetraplegia, paraplegia, parestesia, radiculopathy, and cranial nerve involvement were documented [3,8.34.37]. Morard and Preux reported 80 % of impression of spinal cord in a patient who had pedicular osteochondroma in 6th cervical vertebrae[25]. Wen et al, documented a hereditary exostoses case admitted with sudden tetraplegia and pain[2]. In respective of these reports; it is suggested that the neurological symptoms occur slowly in cervical osteochondromas [14]. Sharma et al reported that neurological symptoms developed between 6 months and 30 years in 10 cases [34]. Ratliff and Voorhees reported a patient who had myelopathy when he was 66 years old [30].

Akagi et al. reported a patient admitted for vertigo and in whom upper cervical osteochondroma was found after examinations [1]. Barros Filho et al, observed an osteochondroma localized in anterior part of vertebrae, which caused dysphagia[4]. George et al. and Kouwenhoven et al, reported cervical osteochondroma cases that had caused headache by vertebral arterial occlusion and vascular compression [15,23]

Although Dahlin supported the rate of 1% and Jaffe claimed it was over 20 %, malignant transformation of osteochondroma was rare [9,13,20]. Green et al. reported sarcamatous change in 2 adult dogs and Fishgrut et al. in a patient[13,19].

In our case, first presenting symptom was solid palpable mass. There was no neurological symptom indicating a spinal cord impression such as myelopathy or radiculopathy or findings of headache, dysphagia and arterial occlusion.

Conventional radiography may not be useful in diagnosis. For instance, Spollone et al. was not sure that CT is useful for determining the border of the tumor. It is characterized by spotted calcification, well-determined border, bone-like density, paraspinal, dumb-bell or exantric intraspinal mass and osteosclerosis at surrounding bone[35]. Morikowa et al. supported that CT is su-

perior to MRI for diagnosis; also Lanzieri et al. advocated that CT is more valuable for determining spinal cord impression than myelography[24,27]. Morikowa et al., Geib and Bridwell supported that CT and MRI should be used in combination for diagnosis; CT is employed for determining the border of lesion and MRI shows the surrounding soft tissue, cartilaginous cap and malign change[14,26]. We used both CT and MRI examination for our patient, and diagnosed osteochondroma with well-defined border. Furthermore, histopathological examination supported our diagnosis.

In asymptomatic cases, there is no need for treatment, but full excision of the tumor prevents recurrence[14]. In cervical cord compression, the only treatment is decompression of neural structures by excision of exostoses[11]. According to Albrecht et al., the rate of neurological recovery is 88-90 % by surgical decompression[2]. Shapiro et al. have suggested that by successful excision, the neurological recovery would be complete[33]. Morard and de Preux reported that in a patient with cervical osteochondroma and spastic walking disorder, by wide decompression, complete recovery occurred[25]. The complications after wide decompression are postlaminectomy kyphosis and vertebral instability [14,21].

According to Bhojraj and Panjwani, kyphosis may be prevented by posterolateral fusion after laminectomy and posterior instrumentation by using Hartshill frame and causing stability[5]. Ergun et al. used anterior plaque for preventing kyphosis after tumor excision[12]. Oga et al, suggested that implantation was dangerous and unnecessary; application of laminoplasty by splitting the spinous process could be adequate for successful fusion [29].

In our case, we found that the osteochondroma was localized at 4th cervical vertebrae and there was no sign of compression of the nerves or spinal canal. For this reason, we planned to perform only tumor excision. However, we found narrowing of intervertebral disc distance between C4 and C5, increasing the intersegmental angle to kyphosis and formation of spontaneous anterior fusion between two vertebrae by the distractive effect of the mass on the lower spinous process. In this condition, we performed posterior fusion autologous grafting between C4 and C5 so as to prevent cervical kyphosis. In postoperative 6th month, we observed solid fusion mass after immobilization with cervical rigid arms for two months.

In conclusion, in this case report we have discussed the treatment and symptoms of a male patient admitted for palpable mass at cervical spinous process, which is a rare occurrence. We have pointed to the formation of the anterior spontaneous fusion by mechanical effect and suggested that in order to prevent kyphosis, performing posterior fusion with excision is an effective option.

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