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A case of monostotic fibrous dysplasia detected in the rib after a sports injury

Spor yaralanması sonrası kaburgada tespit edilen bir monostotik fibröz displazisi olgusu

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A forty-six-year-old man with right lateral chest pain was admitted to our clinic. He had a history of a sports injury in the same region that had occurred while wrestling three months previously. Computed tomography of the chest showed a cystic rib lesion of 6 cm in diameter with calcific inclusion. The lesion was removed with excisional biopsy. The result of the pathological examination was reported as fibrous dysplasia. The patient was uneventfully discharged on the third postoperative day. At the 12-month follow-up, patient was fully recovered and no recurrence was detected. Fibrous dysplasia is a common benign congenital lesion that can involve one or more bones. In this article we present a case of monostotic fibrous dysplasia of the rib which developed after a sports injury. This is the first case of its kind reported in the literature. *Key words:* Fibrous dysplasia; monostotic; sports injury.

Fibrous dysplasia (FD) is the most common benign rib tumor accounting for approximately 30% of all benign tumors of the chest wall.^[1] Approximately 70-80% of cases are monostotic with 20-30% being polyostotic. Six to 20% of monostotic FD occurs in the ribs, most commonly the second rib. Approximately 55% of polyostotic FD has rib involvement, most commonly in the lateral or posterior aspect of the rib.^[1]

It is hard to estimate the true incidence and prevalence of FD, but it is reported to comprise approximately 5-7% of benign bone tumors.^[2] The epidemiology of FD is not well known. The age of patients with monostotic disease ranges from 10 to 70 years old, but recognition is most frequent at 20-30 years of age. The disease does not have gender predilection.^[2]

Herein, we present a case of monostotic FD of the rib as a complication of sports injury. This uncommon complication has not been previously reported in the literature. Kırk altı yaşındaki bir erkek hasta sağ yan göğüs ağrısı ile kliniğimize başvurdu. Hastanın üç ay önce güreş yaparken aynı bölgede incinme öyküsü vardı. Göğüs bilgisayarlı tomografisinde 6 cm çaplı, kalsifik inklüzyonu olan, kistik kaburga lezyonu saptandı. Lezyon eksizyonel biyopsi ile çıkarıldı. Patolojik inceleme sonucu fibröz displazi olarak bildirildi. Hasta ameliyat sonrası 3. günde sorunsuz olarak taburcu edildi. On ikinci aydaki kontrolünde hastalık tamamen düzelmişti ve nüks tespit edilmedi. Fibröz displazi bir veya birden fazla kemiği tutabilen yaygın bir doğuştan lezyondur. Bu yazıda, spor yaralanması sonrası gelişen bir monostotik kaburga fibröz displazisi olgusu sunuldu. Bu olgu literatürde kendi türünde sunulmuş ilk olgudur.

Anahtar sözcükler: Fibröz displazi; monostotik; spor yaralanması.

CASE REPORT

A forty-six-year-old man with severe right lateral chest pain was admitted to our clinic. He had a history of sports injury in the same region as a result of wrestling three months previously. After the injury, a slowgrowing, painful mass was revealed. He had no job which subjected him to a risk of chest injury and had no history of cancer or radiation therapy. Upon physical examination, a 3 cm diameter painful mass was palpated over the angle of the ninth right rib. Chest radiography showed a 5 to 6 cm expansive lesion of the rib with a central radiolucency and a sclerotic border in the posterior ninth right rib. Computed tomography of the chest showed a cystic rib expansion with a diameter of 6 cm featuring calcific inclusion (Figure 1). Complete blood biochemistry was normal, and no other bone lesion was found on a whole body roentgenogram.

The patient was operated for his painful lesion and to establish a definitive diagnosis. The lesion was

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Figure 1. Computed tomography scan of the lesion. A cystic rib expansion of 6 cm in diameter, with calcific inclusion.

completely excised with a 10 cm incision, and the rib was partially resected with surrounding soft tissue. The pathology was reported as a fibrous dysplasia (Figure 2). The patient was discharged uneventfully on the third postoperative day. At the 12-month follow-up, the patient was fully recovered, and no recurrence was detected.

DISCUSSION

Monostotic rib lesions are usually slow-growing and asymptomatic. They may cause pain, soreness, compressive symptoms, and pathologic fractures if they become large.^[1-3]

The basic pathophysiologic process postulated in FD is developmental failure in the remodeling of primitive bone to mature lamellar bone along with a failure of the bone to organize in response to mechanical stress.^[1-3] In addition, the mineralization of the immature matrix is abnormal.

The etiology of the disease has been linked with a mutation in the alpha subunit of the stimulatory G protein ($G_s \alpha$). This gene is located at chromosome 20q13.2-13.3.^[2] It is unclear whether FD has an autosomal dominant or autosomal recessive character.^[3]

Although trauma is not a proven etiologic factor in FD, there have been some reports about cases of FD in which the rib had a history of trauma.^[3-5] McDermott et al.^[3] reported two cases of rib FD with a history of trauma in his series of 11 cases of fibro-osseous lesions of the rib. Nadir et al.^[4] reported one patient with coexisting FD and a bone cyst of a rib after labor trauma. Ferrando et al.^[5] reported a case of post-traumatic costal fibrous dysplasia. Our patient also had a history of sports injury



Figure 2. Pathologic features of fibrous dysplasia. Mixed lamellar and woven bone tissues in the fibroblastic cell proliferation zones were noted (H-E x 100).

from three months previously. However, we have to emphasize again that there is no certain evidence for an interrelationship between trauma and fibrous dysplasia, and the relationships mentioned above may be only be incidental.

Absolute diagnosis is difficult in monostotic FD, and a total excisional biopsy, which can be carried out safely, is necessary for a monostotic rib lesion. Some bone lesions, such as Paget's disease, simple bone cysts, nonossifying fibromas, osteofibrous dysplasia, adamantinoma, and low-grade intramedullary osteosarcoma may suggest FD.^[2]

For rib lesions, total excisional removal of the monostotic lesion is the first choice of treatment in order to establish a definitive diagnosis, prevent complications, such as malignant transformation and pathologic fracture, and relieve compressive symptoms, for example, chest pain, dyspnea, dysphagia, and thoracic outlet syndrome.

The prognosis is generally good in patients with FD.^[2] The natural course of FD depends on the form of the presenting disease and occurence of complications. The prognosis is much better in the monostotic form of the disease.

In conclusion, FD is a common benign tumor of the rib and other bones. Although, it is known as a congenital disease, it may occur after trauma and sports injuries.

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REFERENCES

- 1. Hughes EK, James SL, Butt S, Davies AM, Saifuddin A. Benign primary tumours of the ribs. Clin Radiol 2006;61:314-22.
- 2. DiCaprio MR, Enneking WF. Fibrous dysplasia.

Pathophysiology, evaluation, and treatment. J Bone Joint Surg [Am] 2005;87:1848-64.

- 3. McDermott MB, Kyriakos M, Flanagan FL. Posttraumatic fibro-osseous lesion of rib. Hum Pathol 1999;30:770-80.
- 4. Nadir A, Kaptanoglu M, Goze F. Coexisting fibrous dysplasia and bone cyst of a rib after labour trauma. Eur J Cardiothorac Surg 2005;27:513.
- Ferrando R, Rollandi G, Derchi LE, Quilici P, Tornago S. Post-traumatic costal fibrous dysplasia. A case. Radiol Med 2000;100:279-81. [Abstract]