Giant, multiple intercostal arteriovenous hemangioma

Dev, multipl interkostal arteriyovenöz hemanjiyom

Aydın Şanlı, Ahmet Önen, Sami Karapolat, Banu Gökçen

Department of Thoracic Surgery, Medicine Faculty of Dokuz Eylül University, İzmir

Intercostal arteriovenous hemangiomas are extremely rare tumors. Their reaching large dimensions and being multiple is even more rare. 20-year-old male case, presenting with a swelling on the left anterior thoracic wall, had undergone surgery due to the same condition thirteen years ago. Physical examination revealed three nonpulsatile masses of approximately 15x20 cm, 3x2.5 cm and 5x3.5 cm dimensions. Magnetic resonance imaging demonstrated that they exhibited soft tissue properties. Following surgical resection of the masses, the patient was discharged on the 6th postoperative day. Histopathologically, the lesion was diagnosed as an arteriovenous hemangioma. The patient continues to remain disease-free since 1 year after surgery.

Key words: Hemangioma/surgery; thoracic neoplasms/surgery.

İnterkostal arteriyovenöz hemanjiom çok nadir görülen bir tümördür. Bunların büyük boyutlara ulaşması ve multipl olması ise daha da nadirdir. Sol ön göğüs duvarında şişlik nedeniyle başvuran 20 yaşındaki erkek hasta 13 yıl önce aynı nedenle ameliyat olmuştu. Fizik muayenede 15x20 cm, 3x2.5 cm ve 5x3.5 cm çaplarında, pulsatil olmayan üç kitle saptandı. Manyetik rezonans görüntülemede lezyonun yumuşak doku kitlesi özelliğinde olduğu görüldü. Cerrahi olarak kitlelerin çıkarılmasından sonra hasta altıncı gün taburcu edildi. Histopatolojik tanısı arteriyovenöz hemanjiyom olarak değerlendirildi. Ameliyattan sonra geçen bir yıl süresince hastada nüks gözlenmedi.

Anahtar sözcükler: Hemanjiyom/cerrahi; toraks tümörü/cerrahi.

Hemangioma is a common benign tumor. However, skeletal muscle hemangioma comprises only 0.7% of all benign hemangiomas and among cases with skeletal muscle hemangioma, intercostal musculature location is seen only in 1.4% of the cases.[1] These tumors very rarely reach large dimensions and their being multiple is extremely rare.[2,3]

CASE REPORT

A 20-year-old man was transferred to our clinic for treatment of a tumor on the left side of the anterior chest wall. He had only a progressive swelling in this region. From the history of the patient, we understood that the patient underwent an operation due to the swelling in the same site thirteen years ago, but that a new swelling recurred in the same site four years later and got larger during the last year. Three nonpulsatile masses of approximately 15x20 cm, 3x2.5 cm and 5x3.5 cm dimensions, disappearing with compression were detected during the physical examination. There was no thrill and murmur. A chest roentgenogram showed opacity on the left hemithorax. In the magnetic resonance imaging, a soft tissue mass was detected in the left anterolateral hemithorax (Fig. 1).

In the operation, through an elliptical incision including the thin skin existing on the mass, we reached the tumor and started to resect it from the peripheral tissues. Tumor was attached to the surrounding tissues and these attachments were carefully removed by dissection. Then, we found a giant, well demarcated but not encapsulated soft tissue mass. Right anterolateral thoracotomy was applied. The 5th - 6th intercostal arteries and veins related to the tumor were ligatured. The mass was resected from the thorax in order to remove some latissimus dorsi and serratus anterior muscles after getting rid of vascular contacts individually. The other two small masses were also excised through the same incision. After total excision was performed, the layers were covered appropriately. The patient recovered without any complications and was discharged on the 6th day of the postoperative period. The patient continues to remains well and disease-free since 1 year after surgery.

Received: October 6, 2005 *Accepted:* January 3, 2006

Correspondence: Dr. Sami Karapolat. Dokuz Eylül Üniversitesi Tıp Fakültesi, Göğüs Cerrahisi Anabilim Dalı, 35340 Balçova, İzmir.
Tel: 0232 - 426 69 89 e-mail: samikarapolat@yahoo.com

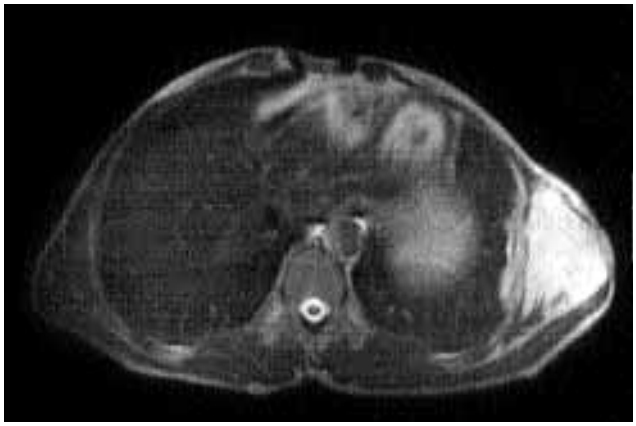

Fig 1. Magnetic resonance images of soft tissue mass on left anterolateral hemithorax.

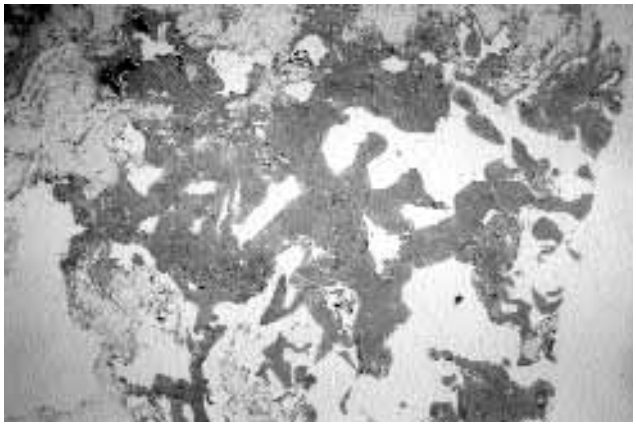

Fig 2. On histopathological examination, the lesion was diagnosed as an intercostal arteriovenous hemangioma (H-E x 2).

The histopathological examination showed that, the small blood vessels infiltrated the surrounding muscle and adipose tissue. The lesion was diagnosed as an arteriovenous hemangioma (Fig. 2).

DISCUSSION

Chest wall tumors account for less than 1% of all tumors and mainly arise in bone and cartilage. Very few of these tumors are vascular. However, intercostal arteriovenous hemangioma is uncommon.[4]

Arteriovenous hemangioma can be divided into two types; those which occur in deep locations with varying degrees of arteriovenous shunting and those occur superficially in the dermis with no significant shunting.[5,6] They are locally invasive tumors and tend to recur if not completely and widely excised.[7]

In general, pulsation and bruits are not recognized because of the sluggish nature of the blood flow. Tumors with large shunting or located close to the skin may have pulsation, thrill or bruit over the mass. Pain is a variable symptom and is a consequence of rapid enlargement and pressure on nearby nerves.[5] The incidence of phlebolith formation is 3-50%. However, our patient was asymptomatic.

Arteriography, magnetic resonance angiography, computed tomography and ultrasonography are beneficial for diagnostic purposes in such type of tumors. However, plain radiography remains the initial method to determine this tumor.[8] We used magnetic resonance imaging without contrast, because we had diagnosed this mass as an intercostal arteriovenous hemangioma on preoperative clinical examination. However, diagnosis of this tumor by magnetic resonance or computed tomography without contrast is difficult. Magnetic resonance angiography is useful method for diagnosis of intercostal arteriovenous hemangioma.

Treatment is difficult, particularly in huge tumors located adjacent to the main vessels and often dangerous. Santiago Recuerda et al.[6] report that, a thoracotomy was performed prior to resection to treat the patient with an intercostal arteriovenous hemangioma. After the resection of mass, ligation of the large, tortuous vessels was performed during thoracotomy. However, despite the ligation, the well-vascularized tumor hemorrhaged severely due to the size and vascular malformation. To avoid this, preoperative arterial chemoembolization is being performed to these patients. We started the operation after obtaining five units of blood, because of the possibility of bleeding and we used three units during operation. In our case, the vascular contacts of the mass were excised after left anterolateral thoracotomy and a larger excision to include a large section of the left latissimus dorsi and serratus anterior muscles. This has been done because complete surgical excision is mandatory to achieve a cure.

Histologically, in intercostal arteriovenous hemangioma, irregular arteries and veins were observed in some areas having close association with one another. This appearance may be misdiagnosed as malignancy, chondroma, and myositis.[9]

In conclusion, this is an extremely rare case because of its originating from multiple focuses, recurrence and large dimensions. This lesion often invades the surrounding muscle. Thus, a complete excision reduces the risk of local recurrence. Currently, in the surgery of intercostal arteriovenous hemangioma, a careful dissection and a wide, complete excision is a very satisfactory method in terms of early and late results.

REFERENCES

1. Ono N, Yokomise H, Inui K, Wada H, Hitomi S. Intercostal hemangioma. Thorac Cardiovasc Surg 1996;44:324-5.

2. Scott JE. Haemangiomata in skeletal muscle. Br J Surg 1957; 44:496-501.
3. Winchester DJ, Victor TA, Fry WA. Intercostal hemangioma presenting as a chest wall tumor. Ann Thorac Surg 1992; 54:145-6.
4. Kubo M, Moriyama S, Nogami T, Kunitomo T, Nawa S. Intercostal hemangioma. Jpn J Thorac Cardiovasc Surg 2004;52:435-8.
5. Tatlis A, De Groot KM, Wainwright H. Intramuscular haemangioma of the chest wall. A case report. S Afr J Surg 1996;34:143-5.
6. Santiago Recuerda A, Corpa Rodriguez ME, Garcia-Sanchez Giron J, Diaz-Agero Alvarez P, Vazquez Pelillo J, Casillas Pajuelo M. Vascular tumors arising in the chest wall: 25 years' experience. Arch Bronconeumol 2005;41:53-6.
7. Mandel L, Surattanont F. Clinical and imaging diagnoses of intramuscular hemangiomas: the wattle sign and case reports. J Oral Maxillofac Surg 2004;62:754-8.
8. Yuan Y, Matsumoto T, Miura G, Tanaka N, Emoto T, Kawamura T, et al. Imaging findings of an intercostal hemangioma. J Thorac Imaging 2002;17:92-5.
9. Fletcher C. Vascular tumours. In: Fletcher C, Unni K, Mertens F, editors. Pathology and genetics of tumours of soft tissue and bone. Lyon: WHO. IARC Pres; 2002. p. 155-78.