

Cervical Paravertebral Osteolipoma: Case Report and Literature Review

Servikal Paravertebral Osteolipoma: Olgu Sunumu ve Literatür Taraması

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Abstract

Osteolipoma is an unusual form of lipoma containing a well-differentiated metaplastic bone. Ossifying lipomas share clinical and radiological characteristics with other benign and malignant tumors; therefore, distinguishing osteolipomas from malignant ones is challenging, especially when using imaging methods. Accordingly, the diagnosing process requires adequate histological characterization to ensure distinguishing it from other neoplasms with a less favorable prognosis, and the surgical excision is considered the most effective treatment. The case we presented was a 61-year-old female, who presented with neck pain without any neurological signs and symptoms. Imaging studies showed a cervical paravertebral ossified lesion treated by total surgical excision and histopathology examination showed osteolipoma. Furthermore, we conducted a literature review that included six cases of cervical osteolipoma. Osteolipomas have a favorable prognosis like conventional lipomas; thus, surgeons should be aware to distinguish ossifying lipomas, which needs a complete surgical resection as the treatment of choice, from malignant lesions included in the differential diagnosis. However, postoperative follow-up is recommended because of relevant clinical information about these tumors.

Key Words: Osteolipoma, Ossifying Lipoma, Atypical Lipoma, Cervical Paravertebral Lipoma, Ossified Lipoma

Öz

Osteolipoma, iyi ayırt edilmiş bir metaplastik kemik içeren alışılmadık bir lipom şeklidir. Kemikleştirici lipomalar klinik ve radyolojik özellikleri diğer iyi huylu ve kötü huylu tümörlerle paylaşırlar. Bu nedenle osteolipomaları kötü huylu olanlardan ayırmak, özellikle görüntüleme yöntemlerini kullanırken zordur. Buna göre, tanı süreci daha az olumlu bir prognoz ile diğer neoplazmlardan ayırt edilmesini sağlamak için yeterli histolojik karakterizasyon gerektirir ve cerrahi eksizyon en etkili tedavi olarak kabul edilir. Altmış bir yaşında, herhangi bir nörolojik belirti ve semptom göstermeden boyun ağrısı gösteren bir kadın olarak sunduğumuz olguda görüntüleme çalışmalarında total cerrahi eksizyon ile tedavi edilen servikal paravertebral kemikleşmiş lezyon saptandı ve histopatoloji incelemesinde osteolipoma saptandı. Ayrıca, altı servikal osteolipoma olgusunun yer aldığı bir literatür taraması yaptık. Osteolipomalar geleneksel lipomlar gibi olumlu bir prognoza sahiptir. Bu nedenle cerrahlar, tercih edilen tedavi olarak tam bir cerrahi rezeksiyona ihtiyaç duyan kemikleştirici lipomları ayırıcı tanıda yer alan kötü huylu lezyonlardan ayırt etme konusunda bilinçlendirilmelidir. Ancak bu tümörlerle ilgili ilgili klinik bilgiler nedeniyle ameliyat sonrası takip önerilmektedir.

Anahtar Kelimeler: Osteolipoma, Kemikleştirici Lipoma, Atipik Lipoma, Servikal Paravertebral Lipoma, Kemikleşmiş Lipoma

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Introduction

The most common repeated soft tissue tumors in the general population are lipomas. Distinguishing these lesions is characterized by the presence of mature fat cells that are surrounded by thin fibrous capsules (1). Grossly, they are yellowish lobulated masses with thin fibrous septal interference strands. Further, they may contain other mesenchymes, resulting in neoplasms, for instance, chondrolipomas, angiolipomas, fibrolipomas, as well as osteolipomas (2).

Although lipomas may be found in any part of the human body, these lesions usually appear in subcutaneous superficial tissues, mostly within the neck, back, and extremities (2) as well as the sternoclavicular region (3). Furthermore, most tumors appear with a similar age in both sexes, in adults aged thirty years or older (3). The rarest and most difficult variant to diagnose is those lipomas that show ossifying changes (4). In a study of 635 cases, less than one percent of lipomas were found to be ossifying (5). They found that the clinical presentation of these lipomas is usually asymptomatic and, thus most cases are not diagnosed for years (6).

If ossification happens on a lipoma outside of the bone, it is referred to as an ossifying lipoma or an osteolipoma. Ossifying lipomas are sharing clinical and radiological characteristics with other benign and malignant tumors (7), thus distinguishing osteolipomas from malignant ones is challenging, especially when using imaging methods (8). Accordingly, the diagnosing process requires adequate histological characterization to ensure distinguishing it from other neoplasms with a less favorable prognosis.

Materials and Methods

We present a case of cervical paravertebral osteolipoma and review the previous reports that concern cervical paravertebral osteolipomas. The search engine which was used in the literature search was PubMed. A combination of these terms was used "osteolipoma" or "ossifying lipoma" or "atypical lipoma" or "Cervical paravertebral lipoma" or "Ossified lipoma". The search was restricted to literature that is related to human beings and the English language. We included six papers published between 2009 and 2016 that conformed to our criteria.

Case Presentation

We illustrate the case of a sixty-year sold female who was diagnosed with diabetes on oral hypoglycemic drugs. The patient had presented to the clinic when she experienced neck pain before around a one-year. There was no family history and no severe previous trauma in the same region. After the physical examination, we found that the patient was neurologically intact. The results of blood tests, including thrombocytes count and coagulability characters, were normal.

Magnetic resonance imaging (MRI) was performed and showed a right-sided mass lesion about 3 X 3.5 X 2.5 cm posterior to C3-C5 hyperintense to isointense to hyperintense on T1, T2 with fat restriction and lobulated wall enhancement with differential diagnosis of liposarcoma, atypical lipoma, and ossifying lipoma (Figure 1).

A computerized tomography (CT) scan was obtained for showing more characterizes. CT revealed a calcified mass lesion



Figure 1a: Magnetic resonance imaging of the right-sided mass posterior to C3-C5

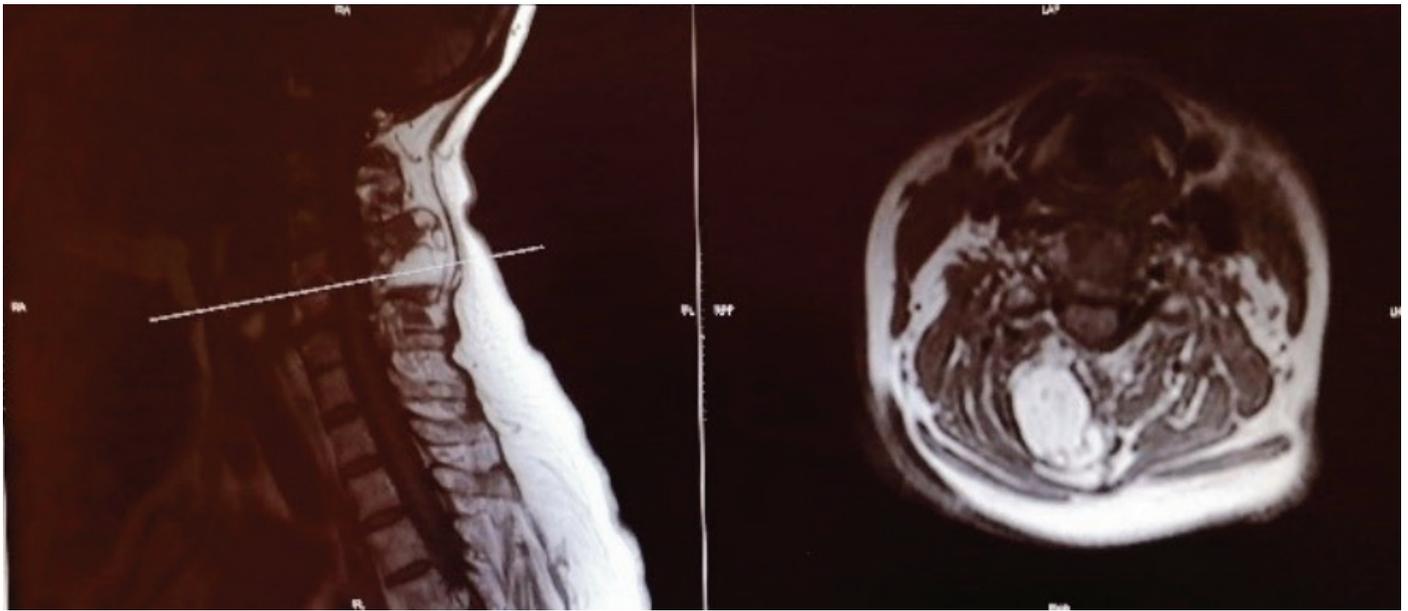


Figure 1b: Magnetic resonance imaging (MRI) of the right-sided mass posterior to C3-C5

right-sided posterior to C3-C5 with thinning of right-sided C4 lamina (Figure 2).

The patient underwent total surgical excision. Intraoperatively, we encountered a white-yellowish non-adherent hard mass underneath the cervical fascia posterior to right-sided C3, C4, and C5 laminae (Figure 3).



Figure 2a: A computerized tomography scan of the right-sided calcified mass posterior to C3-C5

The mass was dissected from the adjacent tissue and excised totally. There were no post-operative complications, so we discharged the patient home on the third day after the surgery. Postoperative MRI and CT scan showed no residual tumor (Figure 4).

Histopathological Findings

Gross examination showed yellow fragmented pieces of fibrofatty to firm tissue with cream/yellow smooth, glistening cut sections, with patchy areas of calcifications and hard bony tissue.

Histopathologic screening showed mature adipose tissue with randomly distributed trabeculae of lamellar bone (Figure 5) and areas with hyaline cartilage undergoing ossification (Figure 6). The adipocytes were monotonous and monomorphic. There was no nuclear atypia, lipoblasts, mitotic activity, or necrosis seen. The case was signed out as osteolipoma.

Discussion

Lipomas are common soft tissue tumors and when these tumors contain a dominant osseous component it is called osteolipoma, and ossifying lipoma if the fat is dominant (9).

A review of 365 cases noted that less than 1% of lipomas were ossified (10). De Castro et al. (5) also reported that there was no osseous metaplasia when histological specimens of 125 facial and oral lipomas were analyzed. Thus, osteolipoma is considered an unusual form of lipoma. They found that the translocations in three osteolipomas were consistent with the karyotypic features of lipoma when cytogenetic analyses of all cases were performed (6).

The mechanism by which lipoma has become ossified is not clear up to now. However, there are two generally accepted theories: first, these tumors may derive from precursor cells that have the capacity to develop into lipoblasts, fibroblasts, and osteoblasts. Second, ossification may also be incited by nutritional deficiency that occurs due to recurrent trauma, metabolic troubles, or ischemia which lead to transform fibroblasts into osteoblasts (1,6,11).

Osteolipomas are usually presented in patients exceeding 45 years with painless mass. Additionally, most cases are asymptomatic and undetected for many years because of the slow growing of this type of tumors (1,11).



Figure 2b: A computerized tomography scan of the right-sided calcified mass posterior to C3-C5

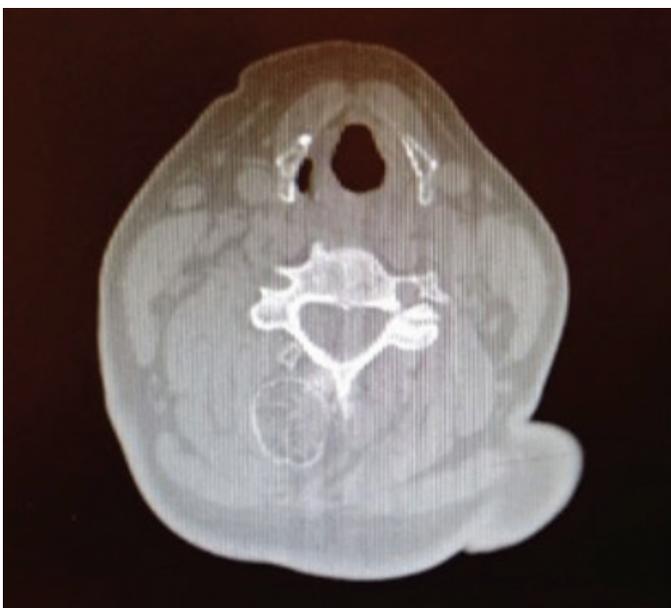


Figure 2c: A computerized tomography scan of the right-sided calcified mass posterior to C3-C5



Figure 2d: A computerized tomography scan of the right-sided calcified mass posterior to C3-C5

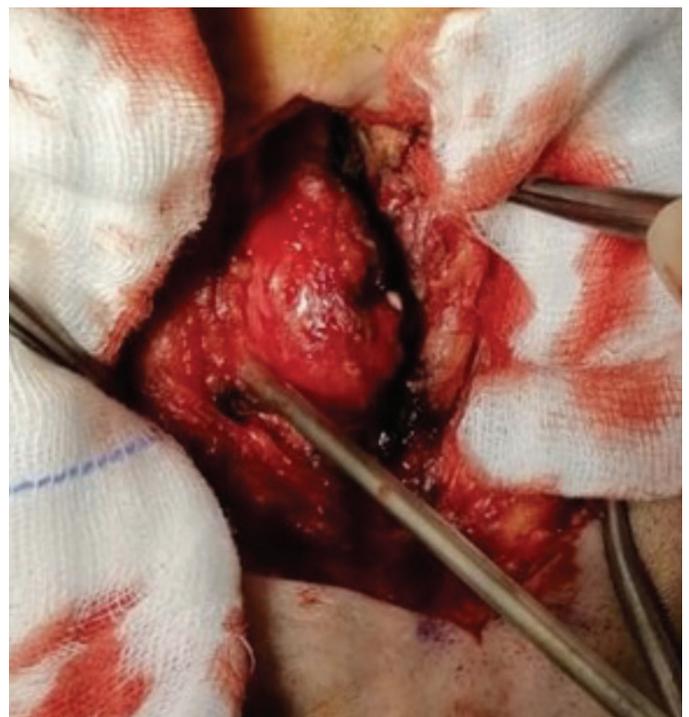


Figure 3: Intraoperative picture of the cervical fascia posterior to right sided C3, C4, and C5 laminae



Figure 4: Postoperative MRI and CT of the Right-Sided mass posterior to C3-C5

MRI: Magnetic resonance imaging, CT: Computed tomography

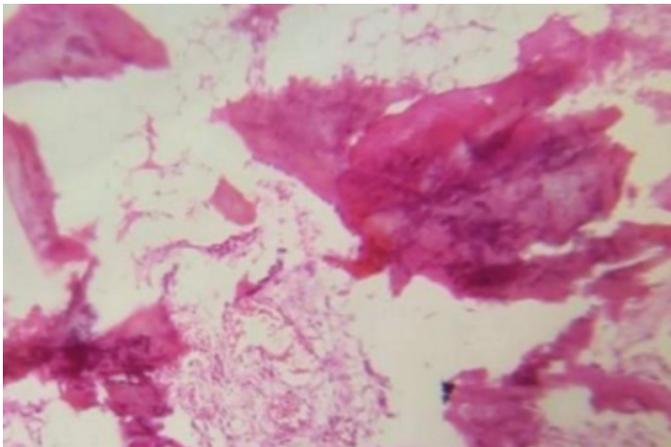


Figure 5: Histopathologic screening showing a mature adipose tissue with randomly distributed trabeculae of lamellar bone

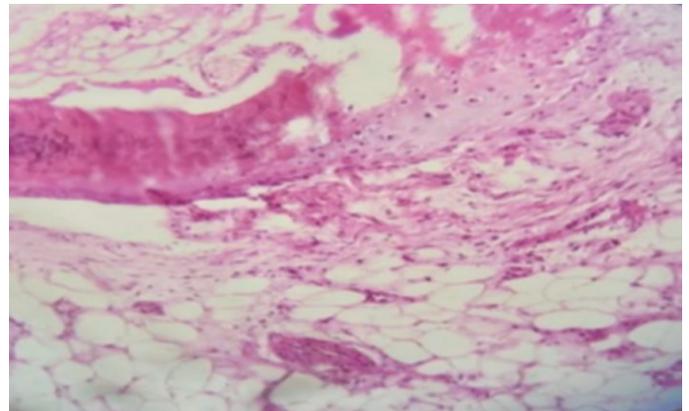


Figure 6: Histopathologic screening of areas with hyaline cartilage undergoing ossification

The differential diagnosis contains various types of other benign tumors such as teratomas, masses with secondary ossification due to trauma, liposarcomas with metaplastic changes, or congenital malformations, as well as surgeons should take consideration into tumor calcinosis, calcification in a bursa, and other conditions such as ossifying fibromas, myositis ossificans, osteosarcomas, and liposarcoma. According to the MRI results of 126 fatty masses, Gaskin and Helms (9) found that osteolipoma may imitate mature liposarcomas, nevertheless they are difficult to distinguish based on imaging alone. However, a Histopathologic test is the only way to give a decisive diagnose and the best treatment would be surgical excision (3,5,7).

Including the current report, a total of seven cases of cervical osteolipoma are included in the literature (Table 1). Five

were female (72%) and the remaining were males (28%), with an age range from 15 to 66 years.

Two of them were intradural (28%), one anterior to C1-C2 (14%) and the remaining four cases were posterior to the cervical spine (58%). While most of the cases were asymptomatic (except neck pain), the intradural lesions presented with dorsal column dysfunction (12) and progressive paraparesis and dysesthesia numbness in both legs (9) with clinical history ranges from 1 to 5 years.

All patients underwent surgical excision, six of them with a posterior approach and an anteriorly located one with the transoral approach. There were no reported adverse outcomes results from the surgeries.

Table 1: Total of seven cases of cervical osteolipoma are included in the literature				
Clinical presentation	Asymptomatic, until a few months ago, when he experienced pain and dizziness.	Asymptomatic	Symptomatic	
Duration (YRS) from diagnosis	2	5	1	
Radiographic appearance	MRI: A mass with a lipid component & calcifications with a possible origin in the right posterior root of C3. CT: Revealed hematic material and small bone spicules with no apparent neoplastic element	MRI: Showed a circumscribed mass compatible with fat & Calcifications	MRI: Midline lesion Posteriorly. CT: Shows the ossific lesion along the spin laminar line near the lamina posteriorly.	
Follow-up	6 months	6 months	24 months	
Attachment/ invasion	Calcified, right-sided, paravertebral mass.	A circumscribed mass, with large calcification.	Severe canal stenosis (4.0 mm) with the compressed cord, with reduced caliber and flattening.	
Approach	Posterior	Posterior	Posterior	
Size (mm)	8x3x4	4x6x3	8 x 9	
Site of the lesion	C2-C6	C2-C6 spinous processes	The cervical spinal canal (C5)	
Age(yrs)/sex	66/M	51/F	61/M	
Article/year	Guirro et al./2015	Yang et al./2013	Aiyer et al./2016	
Clinical presentation	Symptomatic	-	Asymptomatic	Neck pain
Duration (YRS) from diagnosis	2	-	-	1
Radiographic appearance	MRI: At the C6-7 stage, a 3 cm rounded extradural mass with low signal strength and a bright signal portion was demonstrated. The C6-7 VB's posterior scalloping was also discovered. CT: Intraspinal mass that had the density of bone.	MRI: Showed 2 sinus tracts. One of these tracts ended in the bone mass, and the other followed between the laminae of C2 and C3 to the dura. CT: revealed that bone mass, with no direct connection to the axial skeleton.	MRI: No communication between the lesion and C1 or C2. CT: Calcifications	MRI: Mass posterior to c3-c5 hyperintense to isointense on T1, T2 with fat restriction and lobulated wall enhancement. CT: Calcified mass
Follow-up	-	2 years	6 months	-
Attachment/ invasion	No nuclear atypia. No increase cells division rate.	Complete resection in the intradural tract and subtotal resection of the spinal fatty mass.	-	-
Approach	-	Posterior	Transoral	Posterior
Size (cm)	4.5 x 2.5 x 2	7x7	3x2x2	3x3.5x2.5
Site of the lesion	Spinal canal and the dura mater	Posterior cervical spine	Parapharyngeal anterior to c1 and c2	Right paravertebral c3-c5
Age(yrs)/sex	20/f	21m/f	15/f	60/f
Article/year	Lin et al./2009	Brones et al./2009	Bohm et al./2011	Current case/2021

Conclusion

Osteolipoma is an unusual form of lipoma with an asymptomatic clinical presentation. It shares clinical and radiological characteristics with other benign and malignant tumors. These lipomas have a favorable prognosis like conventional lipomas; thus, surgeons should be aware to distinguish ossifying lipomas, which needs a complete surgical resection as the treatment of choice, from malignant lesions included in the differential diagnosis. However, postoperative follow-up is recommended because of relevant clinical information about these tumors.

Ethics

Informed Consent: The manuscript contains no data by which can identify the patient. The informed consent was gained from the patient.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: M.A.A., Y.A., K.A., M.A., H.A., Design: M.A.A., Y.A., K.A., M.A., H.A., Data Collection or Processing: M.A.A., Y.A., K.A., M.A., H.A., Analysis or Interpretation: M.A.A., Y.A., K.A., M.A., H.A., Literature Search: M.A.A., Y.A., K.A., M.A., H.A., Writing: M.A.A., Y.A., K.A., M.A., H.A.

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