Giant cervical lipoma: Case series and literature review

Dev servikal lipom: Olgu serisi ve literatür incelemesi

Ali Bayram¹, Altan Kaya¹, Ebru Akay², Nuri Ünsal¹, Cemil Mutlu¹

¹Department of Otolaryngology, Kayseri Training and Research Hospital, Kayseri, Turkey ²Department of Pathology, Kayseri Training and Research Hospital, Kayseri, Turkey

ABSTRACT

Objectives: This study aims to present a case series comprising giant cervical lipomas with a review of the literature.

Patients and Methods: Of a total of seven patients (4 males, 3 females; mean age 47.4 years; range, 17 to 71 years) operated for giant cervical lipoma in our clinic, age and sex as well as tumor's duration of presence, size, location, and radiological and histopathological findings were recorded. Also, the literature was reviewed for case reports and case series written in English concerning giant cervical lipomas. The terms "giant cervical lipoma" and "giant neck lipoma" were used in PubMed database search.

Results: The main complaint of the patients was a large and painless mass in the neck. In all patients, the mass was removed with total surgical excision without any serious complication and no recurrences were observed in the regular follow-ups. In the literature review, 21 giant cervical lipoma patients were detected in 14 well-documented studies published between January 1973 and June 2017.

Conclusion: Since lipomas usually behave as slow growing, painless and asymptomatic masses, their treatment may be neglected by some patients, causing the masses to reach giant sizes. Management of giant cervical lipomas may be performed without any serious complication with proper preoperative evaluation and meticulous surgical dissection.

Keywords: Lipoma; mass; neck.

Lipomas are benign, slow-growing, mesenchymal neoplasms that are the most common soft tissue tumors of the human body. ^[1] They usually present in the fifth and sixth decades of life and can occur in almost any

ÖZ

Amaç: Bu çalışmada literatür incelemesi eşliğinde dev servikal lipomlardan oluşan bir olgu serisi sunuldu.

Hastalar ve Yöntemler: Kliniğimizde dev servikal lipom nedeniyle ameliyat edilen toplam yedi hastanın (4 erkek, 3 kadın; ort. yaş 47.4 yıl; dağılım, 17-71 yıl) yaş ve cinsiyetleri ile beraber kitlenin bulunma süresi, büyüklüğü, yerleşim yeri, radyolojik ve histopatolojik bulguları kaydedildi. Ayrıca, literatürde dev servikal lipomlar ile ilgili İngilizce yazılmış olgu sunumları ve olgu serileri incelendi. PubMed veri tabanı taramasında "dev servikal lipom" ve "boyunda dev lipom" terimleri kullanıldı.

Bulgular: Hastaların başlıca yakınması boyunda büyük ve ağrısız kitleydi. Bütün hastalarda kitle ciddi bir komplikasyon olmaksızın total cerrahi eksizyon ile çıkarıldı ve düzenli takiplerde nüks izlenmedi. Literatür taramasında, Ocak 1973 ve Haziran 2017 tarihleri arasında yayımlanan, iyi dökümente edilmiş 14 çalışmada 21 dev servikal lipom hastası bulundu.

Sonuç: Lipomlar genellikle yavaş büyüyen, ağrısız ve asemptomatik kitleler şeklinde davrandığından tedavileri kimi hastalar tarafından göz ardı edilebilmekte, bu da kitlelerin dev boyutlara ulaşmasına neden olabilmektedir. Dev servikal lipomların tedavisi uygun ameliyat öncesi değerlendirme ve dikkatli cerrahi diseksiyon ile ciddi bir komplikasyon olmaksızın yapılabilir.

Anahtar sözcükler: Lipom; kitle; boyun.

tissue in the body where fat tissue exists.^[2] Lipomas are mostly seen as small, solitary and asymptomatic lesions that frequently develop on the extremities and trunk.^[3] However, in rare cases, they have a potential

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Correspondence: Ali Bayram, MD. Kayseri Eğitim ve Araştırma Hastanesi Kulak Burun Boğaz Kliniği, 38010 Kayseri, Turkey. Tel: 0352 - 336 88 84 e-mail: dralibayram@gmail.com

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60 KBB Uygulamaları

to demonstrate a gigantic growth, which are called giant lipomas. Sanchez et al. [4] defined giant lipomas as lesions with a size of at least 10 cm in one dimension or weighing a minimum of 1000 grams. In this study, we aimed to present a case series comprising giant cervical lipoma (GCLs) with a review of the literature.

PATIENTS AND METHODS

Medical records of seven patients (4 males, 3 females; mean age 47.4 years; range, 17 to 71 years) who underwent surgery for GCL between January 2013 and June 2017 in the Department of ENT and Head Neck Surgery, Kayseri Training and Research Hospital, Health Sciences University were reviewed including their demographic data, clinical history and presenting symptoms, tumor size and/or weight, location, duration of the disease, imaging and histopathological findings, treatment modalities and follow-up data. The study protocol was approved by the Medicine Faculty of Ercives University Ethics Committee with reference number 2018/258. A written informed consent was obtained from each patient. The study was conducted in accordance with the principles of the Declaration of Helsinki.

A literature review was conducted on GCL for publications written in English on human subjects including single case reports and case series to date. The terms "giant cervical lipoma" and "giant neck lipoma" were used to search online PubMed database. Eligibility criteria of related publications were: (i) case reports or case series of patients with GCL, (ii) giant lipomas defined as lesions with a size of at least 10 cm in one dimension or weighing a minimum of 1000 grams presenting in the neck, [4] and (iii) diagnosis of lipoma

confirmed by histopathological examination. Data were recorded similar to those in our patient group from each paper and pediatric cases were excluded from the study.

RESULTS

Current case series

The clinicopathologic features of the patients with GCL are summarized in Table 1. The main complaint of the patients was a huge and painless mass in the neck (Figure 1a-c). The lesions were present for 12.3 years on average and were growing slowly over time. The patients reported no history of trauma, fever or weight loss and systemic examination was unremarkable. Head-neck examination revealed soft and non-tender neck masses at least 10 cm in diameter that met the criteria of giant lipoma described by Sanchez et al.^[4] Radiological examination of the lesions demonstrated similar characteristics in all patients with classical lipoma except one case of intramuscular lipoma (Figure 2a-d). Neck ultrasonography (USG) of the six patients with classical lipoma showed a neck mass that was isoechoic with subcutaneous fat, whereas computed tomography (CT) revealed a homogeneous and hypodense lesion with no contrast enhancement. In the neck magnetic resonance imaging (MRI) examination, a well-circumscribed, encapsulated and fat-containing lesion with no contrast enhancement was detected that was indicative of a lipoma. In five patients with fine needle aspiration cytology, histopathological findings suggested a lipoma in four patients, whereas it was non-diagnostic for the patient with intramuscular lipoma. Six patients underwent total surgical removal of the tumor via transcervical

	Table 1 Clinicopathological features of case series									
Case	Age/ Gender	Duration (years)	Location	Size (cm)	FNAC	Histopathology	Follow-up (months)			
1	48/M	4	Posterior cervical	20×12×9	Lipoma	Classical	42			
2	17/M	3	Cervicothoracic	14×5.5×4.5	Nondiagnostic	Intramuscular	40			
3	60/M	20	Posterior cervical	17×10×9.5	Lipoma	Classical	34			
4	41/F	4	Posterior cervical	10×4×4	Lipoma	Classical	30			
5	71/F	10	Posterior cervical	15×10×6	-	Classical	24			
6	56/F	15	Posterior cervical	12×6.5×5	Lipoma	Classical	18			
7	40/M	20	Anterior neck	15×9×5	-	Classical	9			
FNAC: Fine needle aspiration cytology.										

approach with the preservation of vital neurovascular structures.

In the patient with intramuscular lipoma, MRI of the neck demonstrated a hyperintense mass equivalent to that of fat on T₁-weighted images and a suspicious infiltration of the lesion into the scalene muscles. The tumor also had a thoracic extension inferiorly below the clavicle and displacing subclavian vascular structures with no sign of invasion according to the CT and MRI examinations. The patient underwent surgery for total tumor removal with a team of otolaryngology, chest and cardiovascular surgery under general anesthesia. During the surgery, the tumor was found to be infiltrated into adjacent muscle structures and these parts underwent frozen section-controlled excision with adequate surgical margins. The thoracic part of the tumor was removed via median sternotomy by chest and cardiovascular surgeons with the preservation of subclavian vascular structures.

The postoperative course was uneventful in all cases and histopathological examination revealed a classical lipoma in six cases (Figure 1d), while the diagnosis was an intramuscular lipoma with clear surgical margins in one case. The patients are free

of tumor with no evidence of tumor recurrence for 28.1 months on average according to the regular follow-ups.

Systematic review of the literature

According to the PubMed database search, we found 19 papers of GCL.[5-22] However, five of the 19 papers were excluded since they did not meet the eligibility criteria.[18-22] Two out of five papers had no data concerning histopathological examination. [18,19] Two more papers were excluded: one due to inadequate information on the size of the lesion^[20] and the other due to smaller tumor size than the defined criteria. [21] The reason for the exclusion of the fifth paper was because of the patient's pediatric age. In the final analysis, we reviewed 14 well-documented papers including 21 patients with GCL published between January 1973 and June 2017.[4-17] The clinicopathological data of the reviewed papers are presented in Table 2. There were 17 male (81%) and four female patients with a mean age of 58 years. The lesions were present for 10.8 years on average in the papers, with the data regarding the duration of the tumor and the main complaint of the patients being a huge painless neck mass. There were also four cases with ulceration of the overlying skin of the tumor. [4,10,12,17] The most frequent tumor site was the



Figure 1. (a) Firm, non-tender mass located in right posterior cervical area; (b) Intraoperative view; (c) Tumor, size 13x9 cm; (d) Histopathological view (H-E×100). Tumor consists of mature adipocytes with small eccentric nuclei. Mature adipocytes exhibit slight variation in cellular shape and size.

62 KBB Uygulamaları

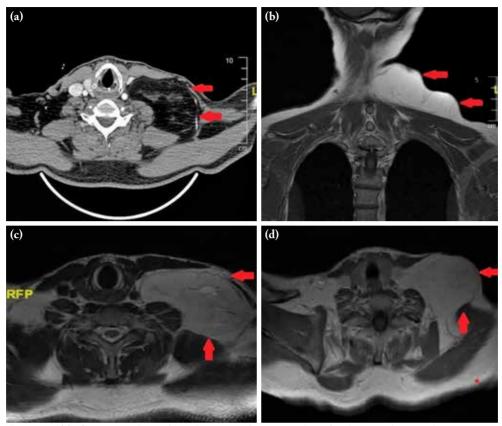


Figure 2. (a) Contrast enhanced axial computed tomography of a lipoma showing homogeneous and hypodense lesion with no contrast enhancement; (b) Coronal T₁-weighted magnetic resonance imaging of a lipoma showing hyperintensity similar to subcutaneous fat signal; (c) Axial T₂-weighted magnetic resonance imaging of a lipoma; (d) Well-circumscribed, encapsulated and fat-containing lesion with no contrast enhancement in axial magnetic resonance imaging with contrast (red arrows show lesion in figures).

posterior cervical region (61.9%) and classical lipoma was the most common histological pattern (76.2%). No recurrence was reported according to the existing follow-up data in the five out of 14 papers. [6-9,11]

DISCUSSION

Lipomas are benign soft tissue tumors that occur in approximately 1% of the population. ^[23] Lipomas can present in any area of the body where adipose tissue exists; however, a head neck lipoma is a relatively rare entity that constitutes less than 15% of all lipomas. ^[3,4] Although solitary lipomas located elsewhere in the body were reported to be more common in females, male predominance has been reported for head neck lipomas. ^[3,24] In the results of the literature review including 21 patients with GCL, the mean age was 58 years with male predilection (81%), while in the present case series, the average age was 47.4 years composed of four male (57.1%) and three female (42.9%) patients.

Solitary lipomas were reported to be present frequently in the fifth and sixth decades of life when the fat tissue begins to accumulate in passive and motionless individuals. ^[2] The data regarding the mean age of tumor presentation in this study were similar to those in solitary lipomas located elsewhere in the body. On the other hand, data concerning the sex distribution of the present case series and literature review revealed a male predominance similar to that reported in the literature for regular head neck lipomas. ^[3,24] Further series including more patients may clarify the accurate age and sex predilection for GCLs.

In the head neck region, lipomas most commonly occur in the posterior neck, nonetheless, cases were also reported in the anterior neck, infratemporal fossa, oral cavity, pharynx, larynx and parotid gland in the literature. Similar to lipomas other than giant tumors, most of the lesions presented in the posterior cervical region (71.4%) both in our study and also in the cases in the literature review (61.9%). Ultrasonography is usually

					Tal	Table 2				
			Clin	icopathologi	cal feature	s of cases in]	Clinicopathological features of cases in literature review			
Author	Year published	Sample (n)	Age (years)	Sex	Duration (years)	Size (cm)	Weight (grams)	Location	Histopathology	Follow-up (months)
Sanchez et al. ^[4]	1993	П	89	M	15	61×27	2,700	Right cervical area	Classical	24
Hirshowitz and Goldan[5]	1973	1	26	M	ı	1	2,450	Posterior cervical	Classical	ı
Copcu and Sivrioglu ^[6]	2005	7	48-63 (56)*	5 M, 2 F	1	20×10×6**	140-460 (245)†	Posterior cervical	Classical	9
Karaçal et al. ^[7]	2006	7	09	ŢŦ	9	12×9	1	Posterior cervical	Pleomorphic	10
Medina et al. ^[8]	2007	7	09	M	15	15×12	ı	Submental	Classical	12
Eryılmaz et. ^[9]	2007	П	72	M	10	42×35	ı	Posterior cervical	Spindle	36
Yakubu et al. ^[10]	2008	Н	70	Ţ	15	30×25×15	ı	Anterior neck	Classical	ı
Derin et al.[11]	2009	Н	57	M	П	17×13×10	262	Left cervical area	Intramuscular	6
Verma et al. ^[12]	2010	Н	89	M	11	22×12	2,200	Posterior cervical	Classical	1
Venkatramani et al.[13]	2010	П	43	M	7	17×11×7	540	Left cervical area	Classical	ı
Basmaci et al. ^[14]	2012	7	54***	2 M	7.5‡	$11\times10^*$	1	Occipitocervical	Classical	1
Singh et al.[15]	2014	Н	62	M	20	38×18×15	ı	Posterior cervical	Fibrolipoma	1
Virk et al. [16]	2016	Н	63	M	1	30×20×20	1,732	Posterior cervical	Spindle	1
Jain et al. ^[17]	2017	Н	70	M	20		2,500	Anterior neck	Classical	ı
* Mean age; ** Largest case; *** Mean age; † Mean weight; ‡ Mean years.	* Mean age;† M	ean weight; ‡ Iv	Iean years.							

64 KBB Uygulamaları

a primary choice among radiological examinations for lipomas in which the lesion typically presents a hyperechoic mass as compared to surrounding muscles. In CT, lipomas often display a homogeneous, hypodense lesion with no contrast enhancement. Lipomas show hyperintensity in T₁-weighted and T₂-weighted fast spin echo MRI sequences, and follow subcutaneous fat signal. Thus, MRI helps to accurately determine exact size, location and extension of the lesion. For giant lipomas of the neck, MRI may provide additional information, particularly in terms of the relationship between the lesion and vital neurovascular structures. In the present study, all patients had proper radiological examinations including MRI that were tasked for diagnosis and meticulous surgical plans.

Histopathologically, lipomas demonstrate a neoplastic lesion with firm fibrous capsule consisting of mature adipocytes arranged in lobules, and may also contain blood vessels, muscle fibers, fibrous septa, and/or areas of necrosis or inflammation.^[1] Histologically, lipomas can be classified as classic lipomas or as variants including atypical, pleomorphic, spindle, intramuscular, myxoid, salivary gland lipomas, angiolipomas and fibrolipomas. [26] Although simple lipomas do not usually pose a diagnostic challenge, a giant neck mass such as a GCL in the head neck region should warn clinicians in terms of a possible malignancy such as liposarcoma.^[18] It has to be emphasized that neither a CT nor a MRI can confidently differentiate lipomas from liposarcomas and histopathological examination is the only way to make a proper distinction. Liposarcomas may resemble lipomas histologically but presentation of scattered atypical fibroblasts and/or signet ring cells in liposarcomas provide diagnostic differentiation.[1] Surgery is clearly the best treatment option for lipomas since a well-defined capsule allows readily complete tumor removal. Different treatment modalities have also been described for lipomas including liposuction^[27] or steroid injection, [28] although none of them has gained popularity. In giant lipomas of the neck, meticulous dissection of the lesion is required so as not to harm vital neurovascular structures due to the close proximity. In the present study, after a proper preoperative clinical and radiological study of the patient, the tumor was totally removed in all cases with no harm to vital neurovascular structures via careful surgical dissection, and histopathological examination revealed a classical lipoma in six cases and intramuscular lipoma in one case. During the surgeries of the six classical GCL patients, we observed that a well-defined fibrous capsule was a major help for the surgeon, regardless of the size of the tumor, and we did not experience any difficulty or complication during transcervical surgical removal of the tumor. In contrast, due to the infiltrative

nature of intramuscular lipomas, total surgical removal may constitute a challenge, particularly in tumors with gigantic growth. In such cases, removal of the infiltrated adjacent structures with the primary lesion has vital importance since these types have a higher rate of recurrence potential due to the infiltrative nature.[11] Frozen sectioning of suspicious infiltrated structures may help to achieve total removal of the lesion with clear surgical margins and also prevent tumor recurrence. Derin et al.[11] reported a giant cervical lipoma invading the carotid artery and total removal of the tumor was achieved by grafting the carotid vessel. In the single case of the present study with intramuscular lipoma, the intrathoracic extension of the lesion necessitated collaboration with chest and cardiovascular surgeons and the tumor was totally removed with no complications. Frozen sectioning was performed to control the surgical margins and the patient has been free of tumor for 40 months. Local recurrence was reported at around 5% for lipomas, mostly related to incomplete excision or infiltrative types. As the recurrence may be seen up to five to 10 years after the first surgery, a close follow-up is mandatory, particularly for intramuscular lipomas.^[29] According to the regular follow-ups of the patients, no recurrence was observed in the present case series including the patient with intramuscular lipoma.

Our study has some limitations. First, we believe that the number of cases with GCL may be much higher than we reviewed in the literature since most of the cases were not reported, or published elsewhere in journals outside PubMed coverage. Second, we did not provide any information concerning the exact frequency of GCLs among the total number of lipomas presenting in the head neck region. Small solitary lipomas are mostly treated in outpatient clinics or secondary medical centers and due to our hospital being a tertiary referral hospital, cases operated for lipomas usually pose either a diagnostic or surgical challenge in our clinic. Third, our study did not produce any information concerning the biological mechanism of the huge size increase of the lesion in patients with GCL. However, in this study, we reported one of the largest case series of GCL with a review of the literature including clinicopathological

In conclusion, since lipomas usually behave as asymptomatic, slow growing and painless masses, treatment of such lesions is neglected by some patients, leading to a huge increase in the size of the tumors. However, management of GCLs can be performed with no serious complications as long as proper preoperative evaluation of the lesion and meticulous surgical dissection are implemented.

Declaration of conflicting interests

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