BENIGN BONE AND SOFT-TISSUE TUMORS OF THE EXTREMITIES CAUSING COMPRESSION OF NERVES

SİNİR BASISI YAPAN BENİGN KEMİK VE YUMUŞAK DOKU TÜMÖRLERİ: CERRAHİ TEDAVİ SONUÇLARIMIZ VE LİTERATÜR DERLEMESİ

Ahmet SALDUZ*' Arda ÇINAR**, Mehmet ALP**, Emre ÖZMEN*, Ömer Naci ERGİN*, İbrahim Levent ERALP*

ABSTRACT

Objective: Compression of nerves by benign bone and soft tissue tumors comprise an uncommon cause of peripheral neuropathies. We aim to present cases of peripheral nerve compression due to benign bone and soft tissue tumors treated in our clinic.

Methods: We report a case series with a total of 16 patients who were treated in our clinic between 2010 and 2015. Mean age of the patients was 28 (2,5-55). Six of the patients had osteochondroma of the fibular head. The remaining 10 patients had various soft tissue tumors localized at different locations. Patients presented with pure sensory, pure motor, or mixed sensory and motor deficits. Mean duration of follow-up was 38 (11-120) months.

Results: All patients underwent surgical excision and were treated additionally with vitamin supplements. All patients regained function within 1 month post-operatively. There was no recurrence at the end of the follow-up.

Conclusions: Compression by tumors should be included in the differential diagnosis and work-up of peripheral neuropathies. Results are excellent with prompt diagnosis and surgical intervention.

Keywords: Benign tumors; nerve compression; neuropathies; osteochondorma.

ÖZET

Amaç: Benign kemik ve yumuşak doku tümörlerinin oluşturduğu sinir basıları nadir bir periferik nöropati sebebidir. Bu çalışmada kliniğimizde benign kemik ve yumuşak doku tümörü nedeniyle oluşmuş periferik nöropatili hastalarda uyguladığımız cerrahi tedavilerin sonuçlarını bildirmeyi amaçladık.

Hastalar ve Metod: 2010 ve 2015 yılları arasında kliniğimizde tedavi edilen benign tümöre bağlı sinir basısı oluşmuş 16 hasta çalışmaya dahil edildi. Ortalama yaşları 28 (2,5- 55) idi. Altı hastada fibular sinire bası yapan fibula başı osteokondromu tespit edildi. Geriye kalan 10 hastada çeşitli yerlerde çeşitli yumuşak doku tümöleri tespit edildi. Hastalar ilk başvuruda pür duyusal, pür motor yada her iki nörolojik deficit ile başvurdu. Ortalama takip süremiz 38 (11-120) ay idi.

Sonuçlar: Tüm hastalar tümörün cerrahi eksizyonu ve ek olarak vitamin takviyesi ile tedavi edildi. Bütün hastalar ameliyat sonrası ortalama 1 ay içerisinde motor ve duyusal fonksiyonu geri kazandılar. Hastaların son takiplerinde nüks izlenmedi.

Çıkarımlar: Periferik nöropati ayırıcı tanısında sinir basısı yapan tümörler unutulmamalıdır. Erken tanı ve uygun cerrahi tedavi ile mükemmel sonuç elde edilebilir.

Anahtar kelimeler: Nöropati; bening tümörler; osteokondroma; sinir basısı.

İstanbul Tıp Fakültesi Dergisi Cilt / Volume: 80 • Sayı / Number: 1 • Yıl/Year: 2017

Date received/Dergiye geldiği tarih: 30.01.2017 - Date accepted/Dergiye kabul edildiği tarih: 25.03.2017

^{*} Istanbul University, Istanbul Faculty of Medicine, Department of Orthopedics and Traumatology,

^{**} Memorial Sisli Hospital, Department of Orthopedics, *** Department of Plastic and Reconstructive Surgery, Istanbul, Turkey

⁽Corresponding author/İletişim kurulacak yazar: ahmetsalduz@gmail.com)

Benign tümörler, sinir basıları

INTRODUCTION

Peripheral nerve entrapments are commonly seen in anatomical tunnels. In the upper extremities, median nerve entrapment in the carpal tunnel and ulnar nerve entrapment in the cubital tunnel are most commonly diagnosed (2, 5). Radial nerve is less frequently involved. In the lower extremities, while peroneal nerve entrapment is seen most frequently, entrapments in the sciatic nerve and all of its 5 terminal branches, namely tibial, deep peroneal, superficial peroneal, sural, saphenous can be seen.

Benign tumors usually present with a painless mass and/or swelling. These lesions may also compress the nerves and cause neuropathic symptoms. Neuropathy at

Table 1. Demographic and clinical features of the patients with their pathological diagnoses and recovery	7
times.	

times.										
No	Age	Sex *	Sensory(S) /motor(M)	Chief complaint	Affected nerve	Location	Diagnosis	Recovery time (week)		
1	35	М	M+S	Hypoesthesia over the deltoid region and weakness in shoulder abduction	Axillary nerve	Axilla	Schwannoma	4		
2	42	М	S	Paresthesia over the distibution of median nerve	Median nerve	Forearm	Schwannoma	12		
3	55	F	M+S	Motor and sensory deficit in radial nerve	Radial nerve	Forearm	Schwannoma	4		
4	25	Μ	M+S	Opposition deficit of the thumb Sensory deficit yazılmalı	Anterior interosse ous nerve	Forearm	Schwannoma	4		
5	45	М	M+S	Extension deficit of the thumb Sensory deficit yazılmalı	Posterior interosse ous nerve	Forearm	Lipoma	3		
6	30	F	S	Paresthesia and pain in the plantar side of the foot	Tibial nerve	Leg	Ganglion cyst	3		
7	44	F	S	Hyperesthesia at the plantar side of the foot and weakness in great toe extension	Tibial nerve	Leg	Ganglion cyst	4		
8	43	F	S	Anesthesia at the plantar side of the foot	Tibial nerve	Leg	Schwannoma	2		
9	34	М	S	Anesthesia at the plantar side of the foot	Tibial nerve	Ankle	Ganglion cyst	4		
10	35	М	S	Sciatalgia	Sciatic nerve	Thigh	Schwannoma	2		
11	2,5	М	М	Drop foot	Fibular nerve	Fibular head	Osteochondroma	12		
12	15	М	М	Drop foot	Fibular nerve	Fibular head	Osteochondroma	1		
13	11	F	М	Drop foot	Fibular nerve	Fibular head	Osteochondroma	3		
14	14	М	М	Drop foot	Fibular nerve	Fibular head	Osteochondroma	2		
15	10	F	М	Drop foot	Fibular nerve	Fibular head	Osteochondroma	1		
16	13	М	M+S	Drop foot	Fibular nerve	Fibular head	Osteochondroma	4		

*: M: Male, F: Female

Benign tumors, nerve compression

the extremities due to compression of nerves by benign soft tissue and bone tumors is nonetheless rare and the literature is sparse for such cases. For that reason, in patients with neuropathic symptoms, other diseases should be considered first, such as spinal pathologies or nerve entrapment due to anatomical structures; while keeping in mind the possibility of nerve entrapment due to a bone or soft-tissue tumor. Due to the necessity for meticulous technique for removing these masses while preserving critical structures, if operative treatment is chosen, the operation itself should be carried out by a specialized team focused on bone and soft tissue tumors.

In the literature there are sporadic case reports or series about benign tumors causing neuropathy (1-4, 7, 8, 10, 12-14). We report a case series with a total of 16 patients. Six of the patients had osteochondroma of the fibular head. The remaining 10 patients had various soft tissue tumors localized at different locations. Of these 10 patients, 6 had schwannoma, 4 had ganglion cyst and 1 had lipoma.

PATIENTS AND METHODS

The Institutional Review Board approved the study. Consents of the patients were obtained for surgeries and publication. Records of the patients presenting with neuropathic symptoms were extracted from the archives. Between 2010-2015, 16 patients with benign tumors who presented with neuropathic symptoms were included in the study. Mean age of the patients was 28 (2,5-55). Six patients had osteochondroma of the fibular head causing fibular nerve compression. The remaining 10 patients had neuropathic symptoms due to soft tissue tumors causing compression of the axillary, median, tibialis posterior, radial, ulnar and sciatic nerve. Their demographic and clinical features are summarized in Table 1.

Six patients (4 male, 2 female), with ages ranging from 2,5 to 15, presented with peroneal neuropathy due to osteochondroma at the fibular head. Physical examination revealed decreased sensation along the peroneal nerve distribution and motor deficits in dorsiflexion and eversion. These patients were initially brought to the hospital by their parents who noticed their typical 'drop-foot gait'. Mean duration of symptoms at the time of diagnosis was 5 (2-12) months. Of the remaining 10 patients with various benign soft tissue tumors, 6 were male and 4 were female. Mean duration of symptoms at the time of diagnosis was 2.4 (1-6) months. Benign tumor was located in the upper extremities in 5 patients and lower extremities in 11 patients. All patients had EMG, Xrays and MRI evaluations before surgery.

Clinical and demographic features of the patients were analyzed with the descriptive statistics.

RESULTS

All patients were treated operatively. Their accompanying medical treatment consisted of vitamin Bs and alfa- lipoic acid until the complete recovery (250 mg B1 vitamin, 250 mg B6 vitamin, 1 mg B12 vitamin and 300 mg alpha-lipoic acid). Nerve function recovered in all patients within the first postoperative month. Mean duration of follow-up was 38 (11-120) months. Schwannomas, with the exception of one case, did not emerge from the affected nerve. They developed from neighboring nerves and caused neuropathic symptoms by compressing the adjacent nerve.

First patient presented with hypoesthesia over the deltoid region along with weakness in shoulder abduction. MRI study showed a mass in the axilla next to the axillary nerve. It was treated with simple excision. Histopathological examination of the tumor confirmed a benign schwannoma (Fig. 1).



Figure 1: 35-yr. old male patient complaining of hypoesthesia on the deltoid muscle. Fig 1a: MRI shows well-circumscribed mass next to the axillary nerve. Fig. 1b: Intraoperative picture of the nerve and the mass with histopathology of schwannoma.

Second patient had a schwannoma in the forearm compressing median nerve and was treated with

excision. The chief complaint was hypoesthesia in the distribution of median nerve (Fig. 2).



Figure 2: 42-yr. old male complaining of hypoesthesia where median nerve dermatome and mass on the forearm. Fig. 2a: MRI shows 3 cm mass on the median nerve trace. Fig. 2b: Intraoperative picture of the mass and excision material.

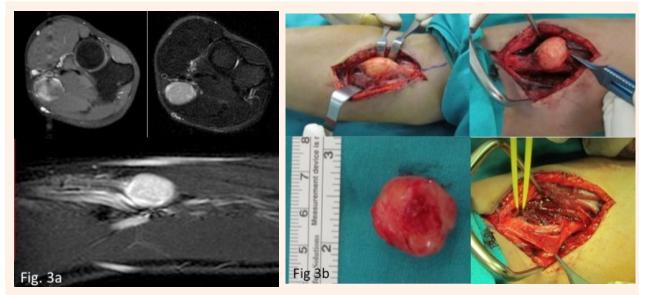


Figure 3a: MRI shows a mass in the antecubital fossa. Fig. 3b: Intraoperative picture of the mass and median nerve.

Following three patients presented with motor deficits in addition to sensory symptoms in the upper extremity due to compression of the radial nerve, anterior interosseous nerve and posterior interosseos nerve respectively. The first one was a 55-years-old female who came to the clinic complaining of progressive motor and sensory deficit of the radial nerve in three months. MRI showed tumoral mass compressing the radial nerve. Excision was performed and histopathological investigation confirmed the diagnosis of schwannoma. Second one was a 25 years old male patient with anterior interosseous nerve compression.

İstanbul Tıp Fakültesi Dergisi Cilt / Volume: 80 • Sayı / Number: 1 • Yıl/Year: 2017

Benign tumors, nerve compression

His presenting symptom was not being able to do opposition while playing the guitar. MRI showed a mass in the antecubital fossa. This schwannoma was excised and pathology investigation confirmed the diagnosis (Fig 3). The third one, a 45-year-old male patient presented with slow-onset paralysis in the right forearm and hand muscles. Electromyographic studies showed severe denervation in the muscles innerved by the posterior interosseous nerve. MRI demonstrated a tumoral mass compressing the nerve. The initial diagnosis was lipoma and the patient underwent surgical excision. Surgical exploration and a biopsy confirmed the diagnosis. Patient had active wrist and finger movements within 3 weeks and returned to his full strength by 6 weeks.

The histopathological diagnosis was ganglion in 3 cases and schwannoma in 1 case (Fig. 4). Presenting symptoms varied from patient to patient, with some patients having anesthesia and some having hyperesthesia. However presenting complaints were all of sensory nature except one patient (Patient 7) who had an additional weakness in great toe extension.



Figure 4. 44-yr. old female complaining of hypoesthesia of the plantar side of the foot and motor deficit of the flexor hallucis longus muscle. Fig. 4a: MRI shows a mass in tarsal tunnel. Fig. 4b: Intraoperative picture of the tumor bed and specimen.

Patient 10 presented with a mass in the posterior thigh with symptoms mimicking sciatalgia. Imaging studies showed a mass next to the sciatic nerve. Tumor was excised and sent for pathological examination, which came back as schwannoma.

Common peroneal nerve was entrapped in 6 children due to solitary osteochondroma of the fibular head. These patients were taken to the hospital due by their parents. These patients underwent several treatments without diagnosis of the primary etiology. Upon initial consultation at our department, osteochondroma at the proximal fibula was detected after physical examination and radiologic assessment. During surgery, the peroneal nerve was dissected, starting from a level above the knee joint. Following nerve release, the osteochondroma was removed, including its cartilage cap (Fig. 5). Consequently, recovery was observed in all six cases after surgery.

DISCUSSION

Clinical manifestations of nerve entrapment secondary to space-occupying masses can be similar to other conventional nerve compression syndromes. Diagnosing these conditions can be difficult and time taking. A complete patient history is of paramount importance. A history of trauma may be a precipitating factor depending on the nerve involved. If indeed a tumor is present, it is unlikely to be felt during physical examination. However, some clues to the presence of a tumor in the physical examination are: hard, nodular feeling on palpation, free motion on the axial plane with restricted motion of the sagittal plane. It is recommended that all nerve compression syndromes be investigated using advanced imaging studies, most commonly with MRI scans. In patients presenting with progressive weakness and sensory changes compatible with a distribution of a nerve, nerve entrapment due to



Figure 5a: Pereoperative X rays shows osteochondroma of the fibular head of the 13 years old male patient presented with fibular nerve dysfunction. Fig. 5b: MRI shows osteochondroma of the fibular head compression to the fibular nerve. Fig. 5c: Treated with nerve decompression and excision of the osteochondroma. Intraoperative picture of the fibular nerve before and after excision. Fig. 5d: Postoperative 8 month x rays of the patient.

benign soft tissue tumors should be kept in mind. Surgery should be performed in a timely manner to restore any loss of function and prevent further possible nerve damage. Recovery of nerve function in these cases is rapid, if surgery is performed before the nerve damage is irreversible. Surgical exploration can be diagnostic as well, since in some cases tumor may not be identified radiologically prior to operation If possible, testing of nerve function should be carried out intraoperatively. We recommend a double- tourniquet technique, or in other terms RIVA. The placement of double tourniquet should not prevent further exploration of the proximal part of the affected nerve if the need arises.

In this series, follow-up of the patients is short-term; however, all patients had the return of affected nerve function in the span of 1 month. Follow-up of the patients are negative for a recurrence so far within 38 months. We believe that with early surgical intervention and appropriate post-operative medical treatment, postoperative one month is a realistic milestone to expect symptom relief and order further imaging and nerve conducting studies if necessary.

Common peroneal nerve is the most commonly seen nerve entrapment syndrome in the lower extremity. In one study, common peroneal nerve was entrapped in 6 children due to solitary osteochondroma of the fibular head (6). Children, especially before school age, usually do not express sensory deficits. Parents are usually the ones to become aware of the situation when a motor deficit develops. Six patients with fibular head osteochondroma in this series were diagnosed late comparing to their adult counterparts - because of this reason. Another reason for late diagnosis is unnecessary and untargeted work-ups such as brain MRI scans. Osteochondromas are easy to miss on the plain X-rays in pediatric population, unless specifically looked for. For reasons explained above, these patients usually present to the hospital with drop foot gait. In this patient group, motor deficits are more common than sensory nerve lesions, which might be explained by the arrangement of the fascicles inside the common peroneal nerve. The motor fascicles run medially, whereas the sensorial fascicles run laterally. The exostosis grows from the bone surface to the periphery, compressing the motor fibers earlier. This puts motor branches within the nerve more prone to injury by a newly forming osteochondroma(6, 9).

Sciatic neuropathy has a wider range of possible etiologies such as tumors, fibrosis, aneurysms pseudo aneurysms and endometriosis (5). One patient in our series had sciatic neuropathy due to schwannoma in this series, which is a rare cause of sciatic neuropathy.

In four patients, tibial nerve or terminal branches was entrapped in the tarsal tunnel. Tibial nerve passes posterior to the medial malleolus and medial to the talus and calcaneus. The tibia forms the anterior wall of the tunnel, the talus and the calcaneus form the lateral wall, and the flexor retinaculum forms the roof. In most patients, the tibial nerve divides into three terminal branches (i.e. medial plantar nerve, lateral plantar nerve, medial calcaneal nerve) within the tarsal tunnel (11).

Benign tumors, nerve compression

Tarsal tunnel syndrome encompasses not only neuropathy of the tibial nerve but also of terminal branches of the this nerve (9).

Nerve entrapment is seen more commonly in the upper extremity. Benign soft tissue tumors are reported as one of nerve entrapment in the literature. Lipoma and ganglion cysts are the two most common reported etiologies in the literature (1-4, 7, 8, 10, 12-14). In this series pathologic diagnosis were schwannoma in 4 cases and lipoma in 1 case for upper extremities.

CONCLUSION

In patients presenting with neuropathic symptoms, benign tumors should be included in the differential diagnosis. Peroneal nerve entrapment by an osteochondroma must be kept in mind in pediatric cases with drop foot gait. Failure to do so may result in unnecessary imaging studies, wasting of hospital and patient resources and most importantly a delay in patients' treatment.

REFERENCES

- 1. Allagui M, Maghrebi S, Touati B, Koubaa M, Hadhri R, Hamdi MF, Abid A. Posterior interosseous nerve syndrome due to intramuscular lipoma. Eur Orthop Traumatol 2014;5:75-9.
- Dailiana ZH, Bougioukli S, Varitimidis S, Kontogeorgakos V, Togia E, Vlychou M, Malizos KN. Tumors and tumor-like lesions mimicking carpal tunnel syndrome. Arch Orthop Trauma Surg 2014;134:139-44.
- 3. El Hyaoui H, Hassoun J, Garch A, Kassimi EH, El Fatimi A. Compression of the posterior interosseous nerve by a deep lipoma. Joint Bone Spine 2014;3:265.
- Fazilleau F, Williams T, Richou J, Sauleau V, Le Nen D. Median Nerve Compression in Carpal Tunnel Caused by a Giant Lipoma. Case Rep Orthop 2014 Article ID 654934, 5 pages.
- Herskovitz S, Scelsa SN, Schaumburg HH. Focal Neuropathies: Nerve Injuries, Entrapments, and other Mononeuropathies in Peripheral neuropathies in clinical practice. Oxford University Press Oxford, City, 2010; Chepter:18, page: 312-346.
- Katirji MB, Wilbourn AJ. Common peroneal mononeuropathy A clinical and electrophysiologic study of 116 lesions. J Neurol 1988;38:1723.
- Kohyama K, Sugiura H, Yamada K, Hyodo I, Kato H, Kamei Y. Posterior interosseous nerve palsy secondary to pigmented villonodular synovitis of the elbow: Case report and review of literature. Orthop Traumatol Surg Res 2013;99: 247-25.
- Martínez-Villén G, Badiola J, Alvarez-Alegret R, Mayayo E. Nerve compression syndromes of the hand and forearm associated with tumours of nonneural origin and tumour-like lesions. JPRAS Open 2014;67: 828-36.
- 9. Pomeroy G, Wilton J, Anthony S. Entrapment Neuropathy About the Foot and Ankle: An Update. J Am Acad Orthop Surg 2015;23:58-66.
- 10. Sakamoto A, Yoshida T, Mitsuyasu H, Iwamoto Y. Lipoma causing posterior interosseous nerve palsy

or superficial radial nerve paraesthesia. J Hand Surg Eur Vol 2011;36:76-7.

- 11. Singh G, Kumar VP. Neuroanatomical basis for the tarsal tunnel syndrome. Foot Ankle Int 2012;33: 513-8.
- 12. Sirrajelhak M, Fnini S, Arssi M, Garch A. [Compression of the superficial branch of the radial nerve by a synovial ganglion of the elbow (about one case and literature review)]. Chir Main 2013;32:255-7.
- 13. Wang B, Zhao Y, Lu A, Chen C. Ulnar nerve deep branch compression by a ganglion: A review of nine cases. Injury 2014;45:1126-30.
- 14. Yazid BM, Tan BC, Das S, Hassan S, Subanesh S. Compression neuropathy of the common peroneal nerve secondary to a ganglion cyst. Clin Ter 2010;162: 549-52.