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Research Study

Neurilemmomas of the Hand—A Review of the Clinical Presentation, Surgical Outcome, and Potential Risk Factors
手部的神經鞘瘤 – 臨床表現,手術結果和潛在危險因素的綜述

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ABSTRACT

Introduction: Neurilemmoma of the hand may not usually present with the classic triad (mass, differential mobility, Tinel's sign). Failure to recognize a neurilemmoma may result in the inadvertent injury of nerves. Preoperative diagnosis is very important and challenging.
Materials and methods: A retrospective study of patients who underwent surgery for neurilemmoma of the hand between 2001 and 2013 was conducted. Their clinical presentation, surgical outcome and potential risk factors were reviewed.
Results: Twenty-eight cases of neurilemmoma arising from digits and hands in 28 patients were retrieved for study. Seventeen were male and 11 were female; the mean age at surgery was 57.1 years old and mean follow-up was 15.4 months. Nineteen lesions were on the volar side and the others were on the dorsal side. Volar digital nerve was the most common site of involvement. All patients presented with a mass, with an average duration of symptoms of 41.1 months; 35.7% had local tenderness. Differential mobility and Tinel's sign could be elicited in 39.3% and 21.4%, respectively. Correct preoperative diagnosis was made only in 25% of cases. At final follow-up, 82.1% of patients was symptom-free. Numbness, local tenderness and hypertrophic scar were reported in 7.1%, 7.1% and 3.6% of patients, respectively. No recurrence and no wound complications were noted. No reoperation was necessary. Multiple lesions and a positive Tinel's sign were associated with less satisfactory outcome and increased risk of postoperative complications as shown by the statistical analysis.
Conclusion: Neurilemmoma should be one of the differential diagnoses when dealing with hand masses. Tinel's sign should be done routinely on examination to facilitate correct preoperative diagnosis. Loupe magnification should be used during operation for optimal surgical outcome.

中 文 摘 要

引言: 手部的神經鞘瘤通常不會顯現出經典的三聯體Triad(突出物mass,差別移動性differential mobility,提內耳氏徵Tinel's sign)。未能識別神經鞘瘤可能導致神經受到傷害。手術前的正確診斷是非常重要和具有挑戰性的。
材料和方法: 我們對由2001年至2013年接受手部神經鞘瘤手術的病人進行回顧性研究。我們回顧了臨床表現,手術結果和潛在危險因素。
結果: 研究包括了共有28例病人,共有28例的手指和手部的神經鞘瘤。手術時病人的平均年齡為57.1歲,平均隨訪時間為15.4個月。當中17人為男性。19個神經鞘瘤位於手掌側,其他位於手背側。手掌側的手指神經是最常見涉及的部位。所有病人均出現平均症狀持續時間為41.1個月的腫塊。35.7%有局部壓痛。差別移動性和提內耳氏徵可分別出現在39.3%和21.4%的病例。在最後的隨訪中,82.1%的病人是無症狀的。麻木,局部壓痛和疤痕肥大分別出現在7.1%,7.1%和3.6%的病人中。研究中未發現有復發和傷口併發症。沒有病人需要重做手術。統計學上發現多發性和陽性的提內耳氏徵都和手術後出現併發症有相關聯。
討論與結論: 在處理手部突出物時,神經鞘瘤應該是鑑別診斷之一。提內耳氏徵的檢查應該在每一個病人身上施行,以便於術前作出正確的診斷。摘除手術Enucleation有著良好的手術結果。

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Introduction

Lumps and bumps in the hand are common presenting features in an orthopaedic clinic. The common differential diagnoses include ganglia, giant cell tumours of the tendon sheath, and haemangioma. Neurilemmomas are occasionally seen. Accurate preoperative diagnosis and management of a neurilemmoma are challenging, as neurological complication is higher because of its close relationship to nerves. Preoperative assessment and investigation in terms of diagnosis and surgical planning such as nerve grafting are important. The typical features of a neurilemmoma are local swelling, positive Tinel's sign, differential passive mobility between longitudinal and transverse planes, and its close relationship to nerves.

Methods

A retrospective study was carried out by recruiting patients who had undergone surgery for neurilemmomas of the hand from 2001 to 2013 in our hospital. All cases were confirmed histologically. Clinical presentation, surgical outcomes, and possible potential risk factors were reviewed. Postoperative numbness, weakness, local tenderness, hypertrophic scar, and recurrence of tumour were studied as well. Short-term complications as well as long-term outcomes were also analysed. All other parameters including age, sex, location of lesion, size of lesion, symptom duration, solitary or multiple lesions, preoperative signs, and symptoms were recorded. Their correlation with long-term complications was analysed using Fisher's exact test and paired *t* test.

Results

Twenty-eight lesions in 28 patients were retrieved for our study. Among these 28 patients, 17 were male and 11 were female. The average age of patients was 57.1 years (range, 22–86 years; Figure 1) and the average follow-up duration was 15.4 months (range, 4–60 months). Solitary lesions were found in 22 patients (78.6%), and the other six patients had neurilemmomas in other areas of the body. For the lesions in the hand, 20 were in the left and eight in the right hand. Nineteen lesions (67.9%) were over the volar side and nine (32.1%) lesions were over the dorsal side. Among the 28 lesions, 14 were in the digits with the middle finger being the commonest. Nine lesions were over the web space, while the remaining five lesions were either in the palm or over the dorsum of the hand. Concerning the origin of the lesions, 14 neurilemmomas originated from the volar digital nerves, followed by seven from dorsal digital nerves and four from the common digital nerve. Two lesions originated from dorsal sensory branches of the radial nerve and one lesion from the main branch of the median nerve in the palm.

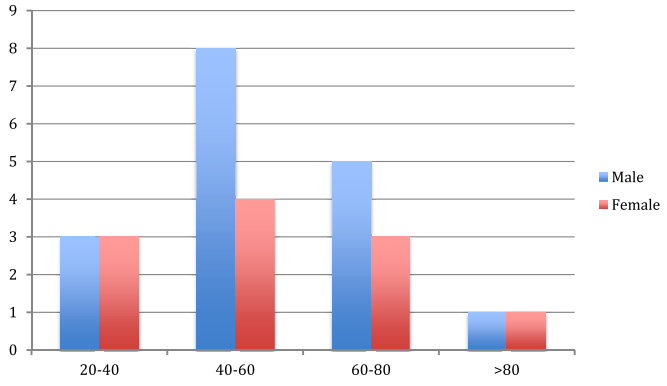

Figure 1. Age and sex distribution of patients.

All patients presented with a mass with an average duration of symptoms of 41.1 months (range, 6–120 months) before the first clinic consultation. Of the patients, 75% reported progression of the size of the mass, while pain and numbness were reported by 17.9% and 10.7%, respectively. There was no complaint about motor weakness. On average, the masses were 15.3 mm in diameter (range, 5–35 mm). Tenderness was noted in 35.7%. Different from neurilemmoma in major peripheral nerve, in which differential mobility and Tinel's sign are common, differential mobility and Tinel's sign were only noted in 39.3% and 21.4%, respectively. Translucency was noted in 35.7%, but it may not be reliable because of the small size of the mass. Correct preoperative diagnosis was made only in 25% of cases. Others were misdiagnosed as ganglia, giant cell tumours of tendon sheath, nonspecific masses, or skin nodules. Magnetic resonance imaging (MRI) investigation was performed in 17.9% of cases mainly involving the palm and volar web space, in order to identify their relationship to deep structures before the operation. All of these cases could be diagnosed with MRI correctly. Correct intraoperative diagnosis could be made only in 67.9% of cases. All patients had the diagnosis confirmed histologically.

Enucleation or excision with preservation of nerve fascicles or suspected nerve fascicles could be achieved intraoperatively in 96.4% of the cases. In one patient with a lesion (35 mm in diameter) arising from the common digital nerve at the fourth web space, the ulnar digital nerve to the ring finger was divided accidentally during tumour resection and repair was done. In the immediate postoperative assessment, numbness of the involved fingers or regions was noted in 35.7% of cases. Among them, 80% had full recovery of sensation at the final follow-up. Permanent paraesthesia was found only in 7.1% (2 patients—1 patient with nerve repair done and another with a tumour arising from the median nerve of the palm just distal to the transverse carpal ligament). Other complications included local tenderness (2 patients, 7.1%), which is likely caused by neuroma formation, and hypertrophic scar (1 patient, 3.6%). In summary, 23 patients (82.1%) were symptom free at the final follow-up consultation. No recurrence was noted, and no second operation was performed. Relationships between long-term complications and various parameters, including age, sex, preoperative symptoms and signs, solitary lesion or multiple lesions, side of limb, size of lesions, and location of lesions, were also analysed. Multiple lesions and a positive Tinel's sign preoperatively were associated with a less satisfactory outcome and a borderline increased risk of postoperative complications as shown by statistics (Table 1).

Discussion

Ozdemir et al[1] and Rockwell et al[2] presented case series of neurilemmomas in the hand. They described the pattern of lesion distribution within the hand similar to our findings, but did not comment on whether the lesions were arising from the volar or the dorsal side. We found that about two-thirds of the lesions were arising from the volar side, which was more common. Digital nerve is the most commonly involved nerve of origin.

A neurilemmoma is a benign peripheral nerve sheath tumour arising from the Schwann cells. They usually present as a slowly progressive painless mass, and some patients may experience pain, and sensory or motor deficit. The classical triad of neurilemmoma is the presence of a mass, positive Tinel's sign, and differential mobility. The diagnosis is highly likely if these signs are present. All patients in the present series presented with a mass in the hand, and 21 (75%) patients had size progression. However, differential

Table 1
Statistical analysis of potential outcome-related factors

Potential outcome-related factors		No. of cases with complications	No. of cases without complications	*p*
Sex	Male	4	13	0.62
	Female	1	10	
Side of limb	Left	5	15	0.28
	Right	0	8	
Multiple versus solitary	Multiple	3	3	0.05*
	Solitary	2	20	
Location of lesion	Volar	5	14	0.14
	Dorsal	0	9	
Tinel's sign	Positive	3	3	0.05*
	Negative	2	20	
Differential mobility	Positive	2	9	>0.99
	Negative	3	14	
Translucency	Positive	0	10	0.13
	Negative	5	13	
		Mean value of cases with complications	Mean value of cases without complications	
Age (y)		57.2	56.6	0.79
Symptom duration (mo)		38.4	53.6	0.63
Size of lesion (mm)		14.5	19.0	0.93

* Exact value is slightly greater than 0.05.

mobility and Tinel's sign were present only in 11 (39.3%) patients and six (21.4%) patients, respectively. Only three patients (10.3%) had the complete triad. In the literature, there was a large variation in the frequency of Tinel's sign in neurilemmomas of the hand (19–71.4%).[1,2] When considering neurilemmomas in all regions, Tinel's sign was a more consistent finding (60–81%).[3,4]

Diagnosing a neurilemmoma of the hand preoperatively is challenging. In our series, correct preoperative diagnosis was made only in seven cases (25%). Other studies had similar yielding rates (19–45.4%).[1–3,5] A neurilemmoma is most commonly misdiagnosed as a ganglion. Phalen[5] commented that neurilemmomas often have a somewhat cyst feel and may occasionally contain small cystic areas. In our series, cystic texture and translucency were noted in 10 cases, and correct diagnosis was made in only one case (10%; Figure 2). The translucency guided the clinician to the diagnosis of ganglion. Daines and Strauch[6] reported two cases of neurilemmoma in the dorsal wrist and volar proximal phalanx; there were positive translucency and preoperative diagnosis of ganglion was made. Histologically, Antoni B areas showed hypocellularity with myxoid degeneration and a high water content, which might account for the signs of translucency.

Rockwell et al[2] commented that the absence of a specific clinic diagnostic test, variable symptoms and signs, and rarity of the lesion contribute to a low rate of preoperative diagnosis. Imaging studies such as ultrasound and MRI may help reach the preoperative diagnosis. In MRI, a neurilemmoma will have intermediate intensity in T1-weighted images and hyperintense signal in T2-weighted images with some heterogeneity. However, neurilemmomas, neurofibromas, and malignant nerve sheath tumours could not be differentiated with certainty using MRI alone.[7] In our study, MRI was performed in five volar lesions and all achieved correct diagnosis preoperatively. In our opinion, MRI may be useful for making the diagnosis preoperatively. However, the cost is very high for its routine use, and it should be reserved for cases with a large mass, with the involvement of the major nerve, when the lesion is suspected to be attached to important deep structures, or when nerve graft is expected.

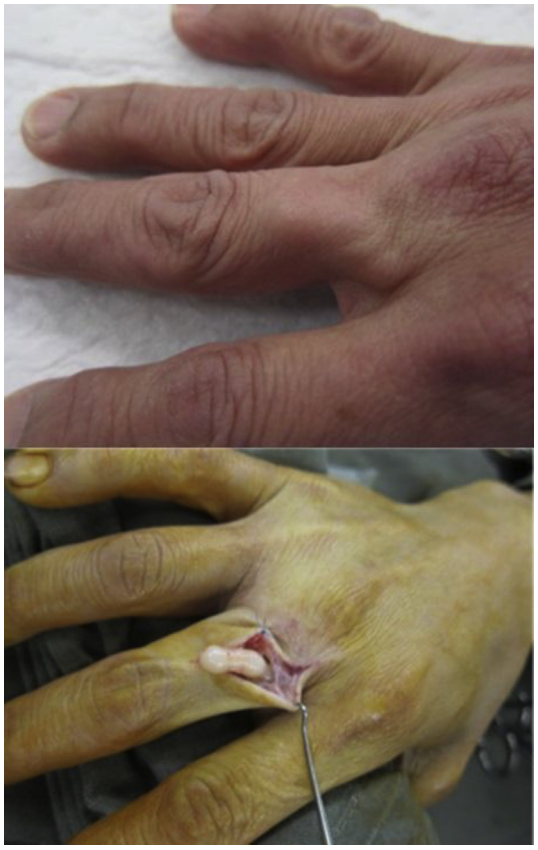

Figure 2. Neurilemmoma at the dorsal side of the right middle finger, with a cystic texture.

Neurilemmoma are closely linked to nerve structures. This characteristic helps in intraoperative diagnosis. Correct intraoperative diagnosis was achieved in 19 out of 28 cases in the present series (67.9%). Loupe magnification or microsurgical technique should be adopted when a neurilemmoma is suspected preoperatively or during operation.

Excision without nerve fibre sacrifice (enucleation) gave good results in our series. Out of 27 cases that underwent enucleation, only one case, in which the lesion arose from the median nerve, has residual numbness. Even with the help of loupe magnification and meticulous surgical techniques, removal of the tumour without damaging any fascicles could still be difficult. Complications still occur due to damage of fascicles along the neurilemmoma and traction injury to nearby fascicles.[8] Various postoperative complication rates at immediate postoperative period and final follow-up were reported (Table 2). Complication rates in our series are comparable with those of other reports.[1,8–11] Although most cases with sensory impairment will recover with time, the patients should be informed about this risk before surgery and it should be included in the informed consent.

In Sawada et al's[12] series of 18 cases of neurilemmomas involving the upper and lower limbs, proximal lesions, lesions larger than 40 mm, lesions with preoperative pain, preoperative sensory deficit, and lack of mobility on examination were poor outcome factors. Park et al[11] also reported that large size was a poor prognostic factor of their 56 upper limb cases, while duration of symptoms and age were not important. Oberle et al[8] also reported that larger diameter and long duration of symptoms were related to less satisfactory surgical results.

We observed that there was a higher complication rate in patients with multiple lesions ($p = 0.05$). A positive Tinel's sign also

Table 2.
Postoperative complications in hand neurilemmoma

	No. of cases	Numbness		All complications		
		Immediate postoperative	Final follow-up	Immediate postoperative	Final follow-up	
Ozdemir et al[1]	14	7 (50)	1 (7.1)	7 (50)	1 (7.1)	
Oberle et al[8]	16	3 (18.8)		5 (31.3)		Data not specified whether from immediate postoperative or final follow-up
Akambi and Dubert[9]	8	3 (37.5)	N/A	3 (37.5)	N/A	
Kehoe et al[10]	79	N/A	N/A	N/A	13 (16.5)	Review cases from all parts of the body
Park et al[11]	56	N/A	N/A	41 (73.2)	17 (30.4)	Review cases in upper limbs
Our series	28	10 (35.7)	2 (7.1)	13 (46.4)	5 (17.9)	

Data are presented as *n* (%).
N/A = not available.

correlates with postoperative complications ($p = 0.05$). Both factors showed borderline statistical significance. The neurophysiological basis of Tinel's sign is presumed to be the lower threshold of regenerating or injured nerves to mechanical stimuli, which permits ectopic generation of orthodromic action potentials.[13] Traction injury and possible damage to the already compromised nerve during dissection may be the causes of a higher complication rate. Other factors were not statistically significant. However, study involving a larger series should be carried out to confirm these outcome factors for definite statistical significance. In addition, our study was a retrospective study, and Tinel's sign may not be tested in every case as the physical examination was not standardised. This would result in an apparent lower sensitivity of Tinel's sign.

Conclusion

A neurilemmoma is a less common but important differential diagnosis of hand masses. Failure to recognize a neurilemmoma may result in inadvertent resection of a nerve. Clinical signs are usually nonspecific; although a positive Tinel's sign would guide the diagnosis, the yielding rate is low. MRI is useful in making the diagnosis, but should be selected for larger masses, preoperative planning, deep structure adhesion, or major nerve involvement. Nerve graft is rarely required, but loupe or microscopic magnification should be used for surgical dissection. Enucleation yields good outcomes in terms of neurological deficit and recurrence.

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Conflicts of interest

The authors have no conflicts of interest to declare.

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