



Ulnar nerve entrapment in Guyon's canal caused by a ganglion cyst: two case reports and review of the literature

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Abstract

In this article, two cases of ulnar nerve neuropathy in Guyon's canal caused by a ganglion cyst are reported. A review of the literature found 73 cases reported in 35 articles of ulnar neuropathy at the wrist secondary to a ganglion cyst. We investigated the pathogenesis of ganglion cysts, which are separated as extraneural and intraneural. At this anatomical area, there are a variety of clinical factors that can also lead to ulnar nerve compression such as trauma, fracture of the hamate's hook, arterial thrombosis, tumors and other anatomical variations. The symptoms may include weakness, paresthesia, numbness and atrophy of hypothenar muscles. Except the careful clinical assessment, magnetic resonance imaging, ultrasound and electrophysiological control are useful tools regarding our differential diagnosis. The treatment of this clinical condition is surgical. We consider that instant surgical decompression is of the essence and enables postoperatively total recovery.

Level of evidence Level III.

Keywords Guyon's canal · Ganglion · Ulnar entrapment · Ulnar neuropathy at wrist

Introduction

Ulnar nerve entrapment or compressive neuropathy is more common at the elbow (cubital tunnel) followed by the wrist (Guyon's canal) [1]. Guyon's canal or ulnar tunnel is a well-investigated anatomical region where ulnar nerve is susceptible to compression [2]. The causative factors of ulnar neuropathy in Guyon's canal include ganglions and tumors, anatomical variations, trauma, idiopathic reasons, vessel malformations or overuse conditions [2–6]. Ganglions manifest themselves predominantly at the dorsal side of the wrist [7]. Ganglion cysts constitute the most frequent cause of ulnar tunnel syndrome [8–11]. Ganglion cysts at the wrist usually stem from tendon sheaths or are related to degenerative arthritis [7]. However, ganglions may also occur within the epineurium of nerves, with mere few reported cases in the literature [12–16]. A widely known theory regarding

the development of ganglions supports their formation due to mucoid degeneration [7]. On the other hand, concerning the generation of intraneural ganglions, Spinner et al. [17] established the articular (synovial) theory which has been widely accepted. There are several case reports concerning the ulnar nerve compression by ganglion cyst at the Guyon's canal, but the clinical manifestation varies according to the area (zone) of the nerve compression by the ganglions cysts. We report two cases of ganglion cysts at the Guyon's canal and a review of the literature concerning the types of ganglions and the clinical course including the different clinical manifestations according to the area (zone) of the nerve compression by the ganglions cysts.

Case reports

Case 1

A 58-year-old right-handed woman visited our hospital's outpatient department, complaining for gradual weakness, numbness and pain of the left hand for about 3-month period. She mentioned no history of trauma or other heavy physical activity. At clinical evaluation, initially we

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Fig. 1 Abduction weakness of the small digit compared with the other hand, because of the impairment of the third palmar interosseous

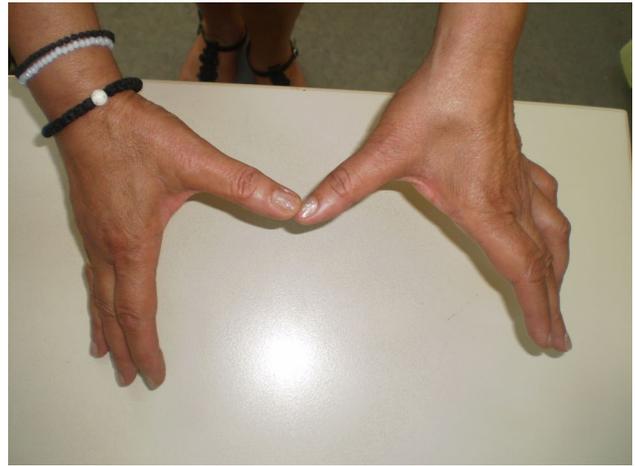


Fig. 3 Atrophy of the first dorsal interosseus



Fig. 2 Ring and small digit clawing. Furthermore, the little finger is positioned abducted because of the dominant action of the extensor digiti minimi (Wartenberg's sign)



Fig. 4 The abduction strength of the small finger was totally retrieved at the final follow-up

observed definite atrophy of the hypothenar, first dorsal interosseous, adductor muscles and ring and small digit clawing (Figs. 1, 2, 3). No palpable mass was found at the volar side of the wrist. Furthermore, during clinical evaluation, Wartenberg's sign and Froment's sign were positive at her left hand. Tinel's sign was also positive at the level of wrist. The hypothenar, first dorsal interosseous and abductor muscles were weaker compared with the other extremity. Moreover, she had sensory deficiency at the volar side of the hypothenar surface. Electrophysiological examination revealed a great decrease in the left ulnar motor amplitude and reduction in the left ulnar sensory amplitude. Moreover, acute and subacute denervation of

the abductor digiti minimi (ADM) and first dorsal interosseous (FDI) were indicated. No alterations in motor and sensory conduction of the ulnar nerve at the level of the elbow were noticed. Magnetic resonance imaging (MRI) revealed a cystic lesion with dimensions 1.4×0.9 cm near the pisiform bone, compressing ulnar nerve at Guyon's canal. Intra-operatively, after an incision over Guyon's canal, the ulnar nerve, ulnar artery and the ganglion were identified. The ganglion was located proximal to the bifurcation of the ulnar nerve, and it was carefully removed. Histological examination confirmed the diagnosis of ganglion cyst. At the postoperative follow-up control, the patient referred to an atypical mild pain at the distribution of ulnar nerve for about 2 weeks. Finally, she retrieved total sensory and motor function in 4 months, according to clinical assessment (Fig. 4).



Fig. 5 The arrow shapes show the atrophy of the first dorsal interosseous and the clawing of the ring and small digits of the left hand

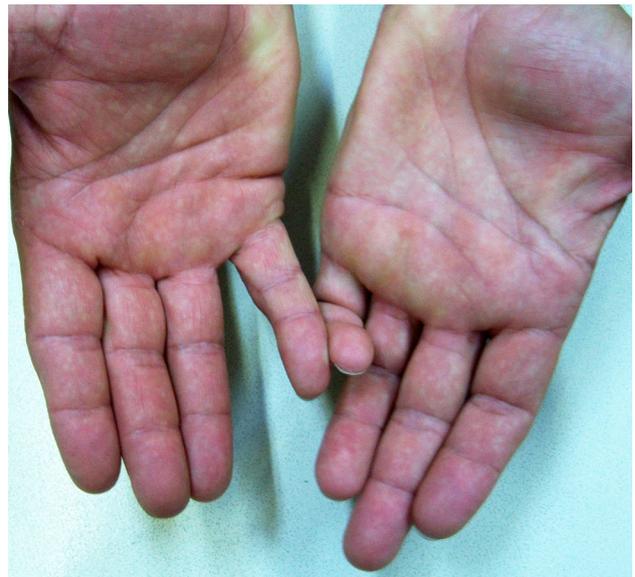


Fig. 7 Abduction weakness of the small digit compared with the other hand, because of the impairment of the third palmar interosseous

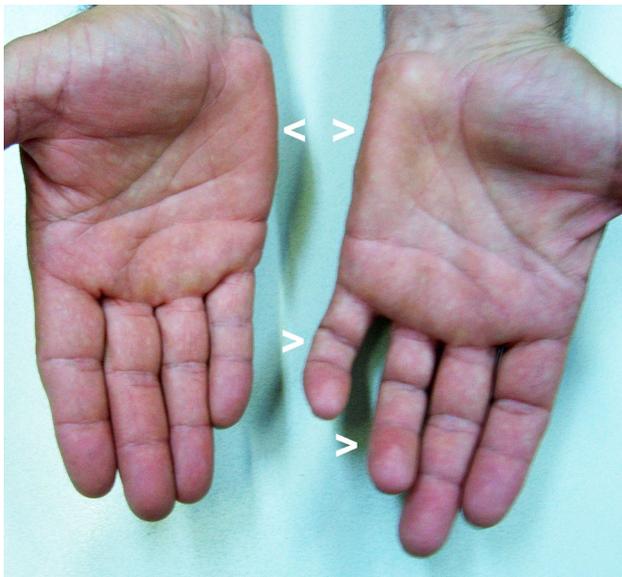


Fig. 6 The arrow shapes indicate the atrophy of the hypothenar eminence and the clawing of the fourth and fifth digits



Fig. 8 The arrow shape shows the atrophy of the first dorsal interosseous

Case 2

A 50-year-old right-handed male patient reported a 6-month history of aggravated pain, paresthesia and weakness of the left hand and wrist. Additionally, the patient complains numbness at the ulnar nerve distribution. On examination, atrophy of the hypothenar, first dorsal interosseous, adductor muscles and ring and small digit clawing were noticed (Figs. 5, 6, 7, 8). Froment's and Wartenberg's signs were also positive, but Tinel's sign was negative. Electrophysiological nerve motor and sensory conduction studies indicated abnormal function of abductor digiti

minimi and of the first dorsal interosseous muscles at the wrist. Three-dimensional, T-2 weighted MRI illustrated a lesion with specific borders and high intensity sign in Guyon's canal affecting the ulnar nerve (Figs. 9, 10). During surgery all the anatomical structures were identified in detail (Fig. 11). The ganglion cyst, which was found proximal to the bifurcation of the ulnar nerve at the wrist, was excised (Fig. 12). Histological analysis verified the diagnosis of a ganglion cyst. On follow-up, no complaint was mentioned and complete clinical recovery was achieved in 4 weeks. The recuperation was evaluated according to clinical signs (Fig. 13).

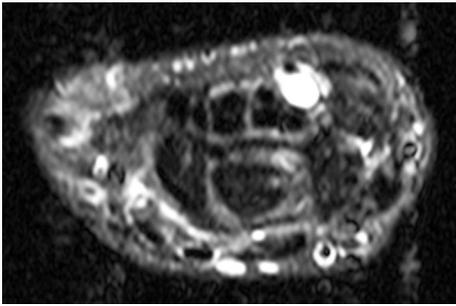


Fig. 9 T-2 weighted MRI, transverse plane. The ganglion cyst is depicted with white color, causing compression at the ulnar nerve

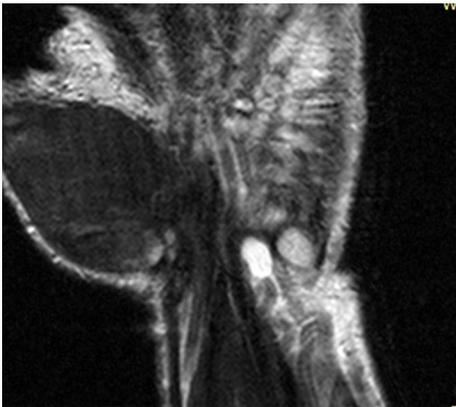


Fig. 10 T-2 weighted MRI, coronal plane. The ganglion cyst is depicted with white color, causing compression at the ulnar nerve

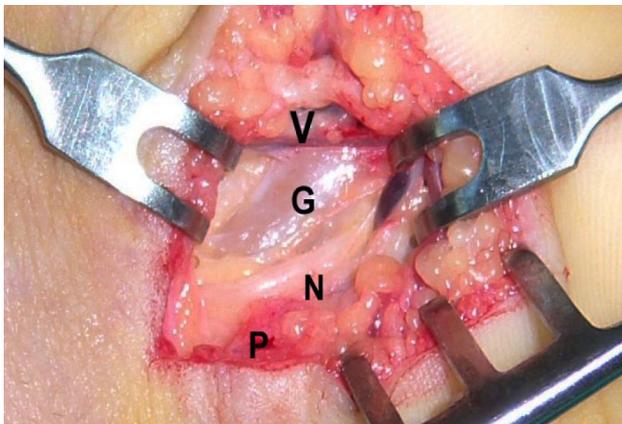


Fig. 11 V: vessel (artery), G: ganglion cyst, N: nerve (ulnar), P: pisiform and pisohamate ligament

Review of literature

We conducted an extensive search of the PubMed/Medline database in the English literature in July and August

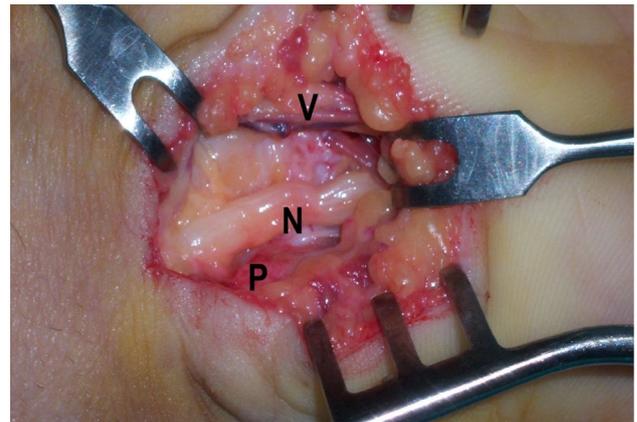


Fig. 12 Surgical exposure of the ganglion cyst. V: vessel (artery), N: nerve (ulnar), P: pisiform and pisohamate ligament



Fig. 13 In this case, the abduction strength of the small finger was retrieved faster compared with the first case, approximately 1 month after surgery

2017. The search terms were “guyon canal,” “ganglion,” “ulnar entrapment” and “ulnar neuropathy at wrist.” An article was deemed eligible if it described ulnar neuropathy at the level of the wrist, caused indispensably by a ganglion cyst. Between 1952 and 2016, we collected 43 reports of ulnar neuropathy at Guyon’s canal caused by a ganglion cyst, but we did not find all the appropriate data in some articles. Totally, we gathered 75 cases reported in 34 articles, including our two cases. The results are demonstrated in Table 1. The statistical analysis was conducted by two authors (ST) and (GD). According to the classification of Gross and Gelberman [51], 32 cases (42.6%) fall into zone I, 41 cases (54.6%) were found at zone II and only 2 cases (2.6%) belong to zone III. Midpalmar ganglions at zone II were observed in 6 cases (8% of all cases). The vast majority were extraneural ganglions (68

Table 1 Cases of ulnar neuropathy in Guyon's canal, caused by a ganglion, reported in the literature

References	Cases	Zone ^a	Age	Sex	Symptoms	DSBS	Side	Treatment	Follow-up ^b
1. Brooks [18]	3	1 all 3 cases	Mean 41	2 Female/1 male	w, par, p, a	6–7 months	2 right/1 left	Surgical excision	4 months–8 year
2. Seddon [19]	4	2 all cases	Mean 45	3 Male/1 female	w, par, p, n	12 months	1 right/3 left	Surgical excision	7 months–16 months
3. Jenkins [20]	1	2	51	Female	w, par, p	2 months	–	Surgical excision	6 months
4. Richmond [21]	12	1 nine cases/2 three cases	Mean 47	2 Male/10 female	w, p, par, n, a	4 months	–	Surgical excision	3 months–12 months/1 case slight weakness 10 years later
5. Vanderpool et al. [22]	13	1 eight cases/2 five cases	43, 54, (two illustrative cases)	1 male/1 female (two illustrative cases)	w, p, par	1–4 months (two illustrative cases)	1 right/1 left (two illustrative cases)	Surgical excision all cases	Few months all cases
6. Hayes et al. [23]	1	2	46	Male	w	½ month	Left	Surgical excision	Partial recovery—4 months
7. Forshell and Hagstrom [24]	2 36 total	2/2 1-20/2-16	66/65	Male/male	p, a, w	25 years/1 year	Left/left	Surgical excision	Relieved by symptoms
8. McDowell and Hencerorth [25]	1	2 Mid-palm	34	Male	a, w	8 months	Right	Surgical excision	14 months No reinnervation
9. Bowers and Doppelt [13]	1	2	50	Male	p, w, a	6 months	Right	Surgical excision	Partly return of strength (5)
10. Kuschner et al. [26]	2	2/2	46/70	2 Male	p, w	9 months/several weeks	2 left	Surgical excision	4 months/several months
11. Feldman et al. [27]	1	2/Mid-palm	45	Female		4 months	Right	Surgical excision	–
12. Shu et al. [28]	1	1	61	Female	w, a	3 months	Right	Surgical excision	Symptoms resolved (24)
13. Kitamura et al. [29]	1	3	44	Female	n, p	Few months	Right	Surgical excision	–
14. Elias et al. [30]	1	2	48	Male	par, p, w	2 months	Right	Surgical excision	Symptoms resolved (1)
15. Kobayashi et al. [10]	1	2/Mid-palm third carpo-metacarpal joint)	71	Female	w, n, p, a	2 months	Left	Surgical excision	2 ½ months
16. Zielinski [14]	1	1	54	Male	w, p, n, a	24 months	Right	Surgical excision	3 months

Table 1 (continued)

References	Cases	Zone ^a	Age	Sex	Symp-toms	DSBS	Side	Treat-ment	Follow-up ^b
17. Nakam-ichi and Tachibana [31]	1 Total 11	1	40	Female (preg-nant)	w, n, a	6 months	Left	Aspira-tion and splint-ing	12 months
18. Papa-thanasiou et al. [32]	1	2	27	Male	a, w	7 days	Right	Surgical exci-sion	1 ½
19. Jacob et al. [33]	1	2/Mid-palm	9	Female	w	Gradually progres-sive	Right	–	–
20. Duggal et al. [34]	1	2/Mid-palm (3rd carpo-meta-carpal joint)	20	Male	w, a	2 months	Right	Surgical exci-sion	Normal motor strength (24)
21. Erkin et al. [35]	1	2	40	Female	P	½ months	Left	Surgical exci-sion	Significant recovery (7)
22. Inaparthy et al. [36]	2	2/2	59/72	male/male	w, a	6 months/2 months	Left/left	Surgical exci-sion	Total recovery (4)/(4 ^{1/2})
23. Chan et al. [37]	1	2	54	Female	w, a	6 months	Right	Surgical exci-sion	Symptoms resolved (4)
24. Chalidis et al. [38]	1	1	52	Female	p, n, a	4 months	Left	Surgical exci-sion	Symptoms totally resolved (30)
25. Pearce et al. [9]	1	1	29	Female	n, w	½ month	Right	Surgical exci-sion	1/3 slight dysesthesia/intact strength
26. Almeida and de Carvalho [39]	1	2	49	Male	w	1 ½ month	Left	Surgical exci-sion	4 months full recovery
27. Kwak et al. [40]	1	1	54	Female	w, par	1 month	Left	Surgical exci-sion	Clinical recovery (1/2)
28. Okada et al. [41]	1	1	51	Male	n, par, a	–	Right	Surgical exci-sion	Symptoms resolved (18)
29. Chen et al. [15]	1 Total 13	1	43	Male	n	12 months	Right	First splint-ing/surgical exci-sion	Symptoms resolved (4)
30. Colbert and Le [42]	1	2	69	Male	w, p	14 months	Right	Surgical exci-sion	10 months total recovery
31. Wang et al. [6]	9	2 all 9 cases	Mean 41	5 Male 4 Female	w	Mean 16 months	6 right 3 left	Surgical exci-sion	Final follow-up total strength recovery

Table 1 (continued)

References	Cases	Zone ^a	Age	Sex	Symp-toms	DSBS	Side	Treat-ment	Follow-up ^b
32. Jayakumar et al. [43]	1	1 dumb-bell shaped/carpal tunnel	59	Male	w, par, p, a	48 months	Dominant	Surgical excision	6 months
33. Bingol et al. [44]	1	3	40	Male	s, n	5 months	Left	Surgical excision	Clinical recovery (2)
34. Gan and Tan [45]	1 total 13	1	51	Female	n, w	8 months	Right	Surgical excision	12 months

w weakness, *par* paresthesia, *n* numbness, *a* atrophy, *s* swelling, *p* pain, *DSBS* Duration of Symptoms Before Surgery

^aGross and Gelbermann et al. [51]

^b(months) when it is not referred

cases—90.6%), and mere 7 cases (9.3%) were intraneural ganglions. The mean age of 64 patients was 47 years with a range of 9–71 year. The incidence of ganglions at the wrist was the same for 50% of male patients (32 cases) and 50% of female patients (32 cases). From 13 reported cases of Vanderpool et al. [22], we included only the two illustrative cases concerning the calculation of the mean age and genders.

As to which is the affected limb, it was mentioned clearly in 49 cases. Right extremity was found with a light predominance in 26 cases (53%), while left extremity in 23 cases (47%). The mean duration of symptoms before the final treatment for 59 cases was approximately 8 months. For the other 16 cases, the exact period was not mentioned. The symptoms included weakness, paresthesia, numbness, atrophy, swelling and pain, and they appeared depending on the location of the ganglion. All patients underwent surgical exploration of the ganglion cyst except one patient reported by Nakamichi and Tachibana [31], who was treated by aspiration of the ganglion and splinting. Finally, at the follow-up examination, the symptoms from 43 cases relieved at a mean period of 8 months.

Discussion

Ganglion cysts are the most frequent soft tissue lesions in the wrist [46]. They present with a light prevalence in female patients [47] and arise approximately 20% from the volar side. They occur usually between the third and sixth decade of life, the mean size is about 3 cm, and they evolve gradually or instantly [47]. Ganglions fall into the category of benign tumors. Yet their pathogenesis still remains a controversial issue among researchers and multiple theories have been proposed in order to explain their generation [7, 46].

In a recent review of the literature, [7, 46] a theory supports that ganglions present with a close connection with adjacent joint. Another assumption concerning their formation is that they are repercussion of synovial herniation or excessive stretch of capsular interface and ligamentous structures. Alternatively, there has been a widespread speculation that ganglions result from mucoid degeneration. On the other hand, intraneural ganglions consist of non-malignant mucinous cysts, which derive from the epineurium of peripheral nerves, and their incidence is infrequent in upper limb [12, 16]. The synovial (unifying) proposal by Spinner et al. [17] has been gaining ground until today among researchers. Moreover, an extensive literature review by Huan Wang et al. [17] validates the hypothesis of the articular origin of intraneural ganglions of upper extremity proportionately to those of lower extremity. Our review includes and investigates both extraneural and intraneural ganglions and refers to their management and follow-up outcomes. Our cases fall into the category of extraneural ganglions.

Guyon's canal is a well-studied anatomical area [48, 49]. Concisely, its margins are transverse carpal ligament at the floor, pisiform bone and the abductor digiti minimi at the medial aspect, at the roof volar carpal ligament and at lateral side the hook of the hamate [2, 48]. Maroukis et al. [17] with an extensive review of the literature scrutinized the progress of our knowledge concerning ulnar tunnel. In 1969, Shea and McClain classified the ulnar nerve compression into three types according to the level of ulnar neuropathy: (a) type I ulnar nerve before its bifurcation, (b) type II deep motor branch and (c) type III superficial branch [50]. In addition to this description, Gross and Gelberman [51] outlined the interdependence between symptoms and pathological occurrence with three anatomical zones of ulnar neuropathy at the wrist. Inevitably, these two last classifications endowed clinicians with understanding and deep knowledge

of ulnar nerve compression at the wrist. We categorized the cases of our review according to these two classifications.

Ulnar tunnel syndrome includes a variety of etiologies which have been described in the literature, especially as case reports [3]. Ganglion cysts, as mentioned above, are the most common cause of ulnar neuropathy at the wrist [3], and they may emerge in all three zones [51]. Moreover, intraneural ganglions manifest themselves more rarely than extraneural ganglions [12]. Midpalm ganglions distal the Guyon's canal are also described in the literature as a rare cause of ulnar deep motor branch entrapment [10, 25, 27]. However, there are other tissue masses such as lipomas [52], giant cell tumors [53], schwannoma, neuroma and osteoid osteoma [5]. Another equal important causes of ulnar neuropathy are anatomical deformities, trauma and fractures, diseases of adjacent vessels, carpal tunnel syndrome, hypothenar hammer syndrome, metabolic pathological conditions or iatrogenic damage [2, 3, 5, 54, 55].

Patients with Guyon's canal syndrome present themselves more often than not with discernible numbness, weakness of grip strength, hypothenar muscles atrophy and pain of the ulnar aspect of the palm [3]. On clinical examination, both upper limbs should be checked comparatively for possible swelling, signs of infection, mobility of all joints, palpable distal arteries or potential neuropathy of radial, median and ulnar nerve [2, 3].

Although a diligent clinical assessment is of the essence, clinicians should consider some extra examinations for a precise diagnosis. X-rays and CT disclose useful information for the condition of the carpal bones, distal ulna and radius and metacarpals (e.g., fractures, displacements, tumors) [54, 55]. Ultrasound is a convenient, cost-effective and patient friendly examination [56, 57]. Sonographic traits of nerve pathology may comprise adjacent soft tissue edema and hyperemia, magnification and alteration in the form of the nerve and hypoechoic swelling [56, 57]. Electrodiagnostic tests are valuable tools regarding the function of nerves. Electromyography and motor and sensory conduction velocities indicate the exact level of the disruption of ulnar nerve [58]. By all means, magnetic resonance imaging (MRI) is the gold standard examination. It depicts in detail all the anatomical lesions and their interaction. Thus, it provides useful information about the pathophysiology of the nerve entrapment and the features of the possible causative factor. Obviously, it plays cardinal role for operative or conservative management [55, 59].

The recommended treatment for ganglion cysts is surgical excision [3, 16]. Presumably, the earlier the surgical decompression of the ulnar nerve, the better the postoperative outcome concerning the motor and sensory recovery. However, other conservative methods have been described in treating ganglions like aspiration [60], aspiration and cyst wall puncture [60, 61] and ultrasonographically assisted aspiration

and splinting [31]. At a European handguide study [62], 35 orthopaedic surgeons who are upper limb experts met with consensus about description, symptoms and diagnosis of Guyon's canal syndrome. Additionally, splinting and surgery were the suggested options of treatment depending on severity and duration of the syndrome, previous treatment and causative factors. In our review, all patients underwent surgical excision of the ganglion cyst and only one patient was treated conservatively with aspiration of the cyst and splinting.

Conclusion

Taking all into consideration, ulnar nerve compression by a ganglion cyst in Guyon's canal is an infrequent clinical condition and clinicians should consider it as a possible cause of ulnar neuropathy at the wrist. It should be noticed that careful clinical evaluation, deep knowledge of anatomy and the appropriate examinations constitute a cardinal role in order to make a definitive diagnosis and determine the right treatment. We believe that early decompression with surgical excision of the ganglion can achieve postoperatively satisfactory recovery.

Compliance with ethical standards

Conflict of interest Authors Stylianos Tottas, Ioannis Kougioumtzis, Zafeiria Titsi, Athanasios Ververidis, Konstantinos Tilkeridis and Georgios I. Drosos declare that they have no conflict of interest.

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