

Tophaceous gout as an unusual cause of carpal tunnel syndrome. A case report

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Case report

Plastic Surgery



Introduction: The carpal tunnel syndrome (CTS) is the most common of the entrapment neuropathies in which the body's peripheral nerves are compressed. It is caused by excessive pressure on the median nerve. Gouty tophi is a rare cause of CTS, with an incidence of 0.6%. Gouty may lead to CTS due to: synovial hypertrophy, tenosynovitis, tophaceous nodules and crystal deposits in nerves, muscles or tendons.

Case report: 80 years old male with a history of a mass in the wrist of the right hand, of 5 years of evolution, which 2 years prior intervention debuts with carpal tunnel syndrome, treated with surgical removal of the mass, with preservation of flexor tendons and medial nerve. Histopathological reported xanthogranulomatous reaction foreign body type and tophaceous gout, 2 years after treatment with good evolution and no symptom recurrence.

Discussion: Gout it's a type of crystal- induced arthritis. Sodium urate crystals with localized inflammation and neutrophil activation that further activate macrophages forming an inflammatory loop. Gout is an uncommon cause of carpal tunnel syndrome. One of the firsts reported cases dated to 1966 by Phalen et al. reported 15 hands with gout in 2469 cases of carpal tunnel syndrome (0.6%). Patil et al. reported three cases of gout among 3216 cases of carpal syndrome (0.9%). Surgical treatment is considered effective for chronic patients.

Conclusions: Gouty tophus is a rare cause of carpal tunnel syndrome; an early diagnosis and control of the gout are necessary to avoid irreversible nerve injury. Delays in the diagnosis and treatment can lead to functional damage and residual deformity. Surgical treatment combined with medical treatment can improve the treatment outcome of gouty tophus.

Key words: Tophaceous gout, median nerve, carpal tunnel syndrome, hand surgery.

The carpal tunnel is a narrow passageway by bones and ligaments on the volar side of the hand, that contains the median nerves and tendons (1). The carpal tunnel syndrome (CTS) is the most common and widely known of the entrapment neuropathies in which the body's peripheral nerves are compressed. It is caused by excessive pressure on the median nerve. If not treated properly, the nerve can be damaged; and lead to numbness, tingling, and weakness in the hand and arm. Causes of (CTS) includes inflammatory diseases, microtraumas, hemorrhages after injury, metabolic, or more frequently idiopathic processes. (2). Gouty tophi is a rare cause of CTS, with an incidence of 0.6% (3). Gouty may lead to CTS due to: synovial hypertrophy, tenosynovitis, tophaceous nodules, crystal deposits in nerves, muscles, or tendons (4). Their appearance in the flexor sheath for the tendons its extremely rare, and may compress the median nerve leading to CTS. (5). Management options for CTS includes,

physiotherapy, steroids oral or injected locally, splitting, and surgical decompression (6).

We present the case of an eighty years-old male with a history of a mass in the wrist of the right hand, of 5 years of evolution, which 2 years prior intervention debuts with carpal tunnel syndrome, treated with surgical removal of the mass, with preservation of flexor tendons and medial nerve. Histopathological report xanthogranulomatous reaction foreign body type and tophaceous gout, two years after treatment with good evolution and no symptom recurrence.

Case report

Eighty years-old male, by profession construction worker, with past medical history of 8 years of gouty arthritis, with no treatment or control. The patient had 3 acute gouty attacks; he took NSAIDs to treat the acute gouty attacks. Previous



Figure 1. Dissection of the right carpal tunnel: mass located in the second and third muscle sheets prior the carpal tunnel, annular ligament and was pressing the median nerve.

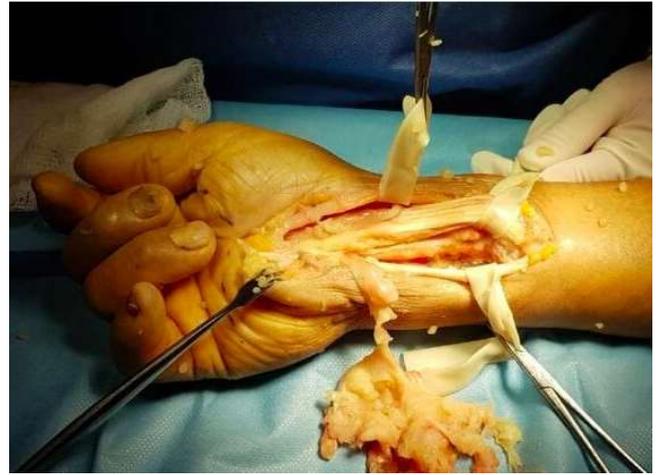


Figure 2. Neurolysis of the median nerve and tenolysis of the adherent tissue.

history of umbilical and inguinal plasty. He refers the appearance of a mass in the right hand at the level of the wrist in volar region that gradually increases size, of 5 years of evolution, 24 months prior to surgical treatment initiates with numbness, and tingling in the thumb, the index, middle finger and half of the ring finger of the right hand, associated with pain and difficulties of movement at the extension and flexion of the hand. On local examination, a palpable hardened mass was circumscribed firm and located on the volar aspect of the distal forearm. It was 6 x 4 cm, elevated, superficial, external mass. The boundary shape was circular and the surface had no tenderness, inflammation, or ulceration clinically. It was movable following the movement of the tendon gliding, the proximal interphalangeal joint of the right of the right index was swollen, with normal hand function and reduced grip strength. Neurological examination revealed numbness in the thumb, index finger middle finger, half of the ring finger, and his right palmar. Force decreased 3 out of 5. Tinel's sign was positive at the wrist. Phalen's maneuver and carpal compression tests were also positive. Electromyography (EMG)

showed the conduction velocity of the median nerve decreased, and the patient sensory function of the median nerve was impaired. The uric acid level was of $727 \mu\text{mol/l}$ ($237.9\text{-}356.9 \mu\text{mol/l}$), and the diagnosis of hyperuricemia was made. The patient was sent to the rheumatology department who initiated medical treatment with febuxostat and allopurinol. Due to the clinical presentation of the mass, the patient is scheduled for surgical exploration of the right carpal tunnel. A longitudinal incision was made on the volar aspect of the distal forearm with the position of fingers in flexion; the mass was located at an unusual site, in the second and third muscle sheets prior to the carpal tunnel, the annular ligament, and was pressing the median nerve (figure 1). The tophaceous gout entered the carpal tunnel when patient middle fingers were completely extended. We remove partially the gouty tophus and preserve integrity of the flexor tendon which was infiltration with chalky with substances. Neurolysis of the median nerve was made completely, and tenolysis of the adherent tissue was released (figure 2). Surgical procedure was carried out without any complications. Three weeks later the histological

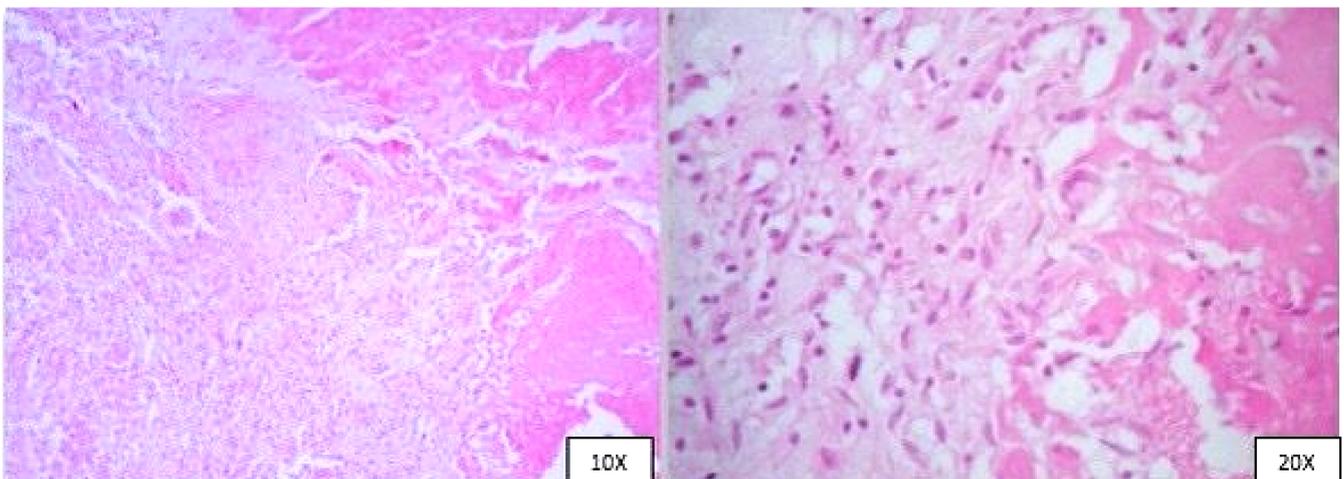


Figure 3. Amorphous substances of various sizes with weak eosinophilic-to-basophilic staining, surrounding aggregates of multinucleated giant cells caused by xanthogranulomatous reaction to foreign body reaction, consistent with tophaceous gout.

reports tissue of ovoid form of 5x3 cm of diameter, anatomopathological diagnosis of xanthogranulomatous reaction foreign body type consistent with tophaceous gout (figure 3). One week after surgery, the patient was able to perform full motions of the operated hand. There was an improvement in the flexor-extension movement; the patient was able to reintegrate to his labor in 3 weeks. 3 months later, symptoms of numbness disappeared, and the fingers range of motion recovered to normal levels. The post-surgical electromyography showed normal nerve conduction velocity 6 months after the surgery. The patient was followed up in the consult every 2 months for another 2 years with no symptom of recurrence. Tendon adhesions, acute attacks of gout did not occur.

Discussion

Gout is a type of crystal- induced arthritis. Sodium urate crystals promote the production of inflammatory cytokines via nucleotide-binding oligomerization domain-like receptor protein 3 (NLRP3) of macrophages leading to localized inflammation and neutrophil activation. Activated neutrophils further activate macrophages forming an inflammatory loop. Gouty tophi are formed when sodium urate sodium urate is trapped by neutrophil extracellular traps (NETs) (7). Decreased collagen and proteoglycan content in the joint along with increased cartilage degradation products in the joint fluid lowers the urate solubility and promotes the formation of monosodium urate crystals (8). Gouty tophi are reportedly common in patients with serum urate levels of > 8.5 mg/dl over a duration > 10 year (9). Gout is an uncommon cause of carpal tunnel syndrome. One of the first reported cases dated to 1966 by Phalen et al. reported 15 hands with gout in 2469 cases of carpal tunnel syndrome (0.6%) (10). Patil et al. reported three cases of gout among 3216 cases of carpal syndrome (0.9%) (11). Carpal tunnel syndrome induced by tophaceous gout has been associated with flexor tenosynovitis in the carpal canal. The tophi can be deposited in and can cause symptoms in various structures including the flexor tendons, tendons sheaths, carpal tunnel floor, transverse carpal ligament, and even the median nerve (12). Medical management of gout should ideally precede, if not prevent, the need for surgical management; initially includes lifestyle and medication changes. Patients should decrease consumption of meats high in purine content, high fructose corn syrup, and alcohol, especially beer (13). Urate elevation medications, such as thiazide and loop diuretics, niacin, and calcineurin inhibitors, should be decreased if possible. Acute gouty attacks can be differentiated based on severity of symptoms and

number of joints involved. If pain is less than 6/10 and gout involves one to two joints, monotherapy can be initiated with (NSAIDs), systemic corticosteroids, or colchicine. Long term management of gout should be initiated when the patient has tophi, multiple gouty attacks per year, or history of uric acid urolithiasis. Initial therapy may include xanthine oxidase inhibitors (allopurinol or febuxostat) (14). Surgical treatment is considered effective for chronic patients. Complete surgical resection its difficult in diffuse lesions; however, by using pharmacotherapy as an adjunct, good results can be obtained. Contrarily, surgical complications, including wound dehiscence, post-operative gout exacerbation, and tendon rupture, have also been reported (15).

It is important to distinguish patients with carpal tunnel syndrome related to gout from those with compressive neuropathy due to thickening of the transverse carpal ligament alone, because subsequent surgical management is different.

In our patient the surgical treatment was successful; partial removal of the gouty tophus and retain of the integrity of the tendons had been achieved with a good result in our patient.

Conclusion

Despite the fact that gouty tophus is a rare cause of carpal tunnel syndrome; an early diagnosis and control of the gout are necessary to avoid irreversible nerve injury. Delays in the diagnosis and treatment can lead to functional damage and residual deformity. Surgical treatment with combined medical treatment can improve the treatment outcome of gouty tophus.

Conflicts of interests

There was no conflict of interest during the study.

Acknowledgements

We want to thank to the department of investigation in our hospital, for assisting us in this labor.

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