CHALLENGING DIAGNOSIS FROM PARONYCHIA TO GLOMUS TUMOR

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ABSTRAK

Tumor glomus, juga dikenal sebagai angioneuromioma, adalah neoplasma jinak yang muncul dari badan glomus, suatu struktur yang berfungsi dalam pengaturan suhu tubuh. Tumor jinak yang langka ini telah dilaporkan di sejumlah lokasi di tubuh, paling sering terlihat di area subungual jari-jari tangan. Hanya sedikit yang telah dilaporkan di daerah kaki, terutama jempol kaki. Trias klasik berupa nyeri spontan, nyeri tekan, dan hipersensitivitas dingin umumnya ditemukan sebagai presentasi klinis dari jenis tumor ini. Seorang wanita berusia 33 tahun datang ke rumah sakit kami dengan keluhan nyeri kronis dan sensitivitas suhu dingin, di jempol kaki kanannya dengan kesulitan memakai sepatu selama 9 tahun terakhir. Hasil rontgen dan pemeriksaan laboratorium normal. Pemeriksaan lebih lanjut dengan pencitraan resonansi magnetik dilakukan dan menunjukkan lesi jaringan lunak berbatas teratur dengan hipointensitas di T1 dan hiperintensitas di T2. Pengangkatan lesi dilakukan dengan eksisi dan laporan histopatologi mengonfirmasi diagnosis tumor glomus pada jempol kaki. Tindak lanjut di klinik rawat jalan menunjukkan bahwa pasien telah bebas nyeri selama 5 minggu setelah operasi. Tumor glomus sebagai kemungkinan penyebab nyeri kronis pada jari kaki sering kali terlambat didiagnosis dan salah didiagnosis sebagai paronikia karena jarang muncul secara klinis, terutama pada jempol kaki. Tujuan dari laporan kasus ini adalah untuk meningkatkan kesadaran dokter dan ahli bedah tentang tumor ini sebagai kemungkinan diagnosis.

Kata kunci : hipertensi, kebidanan komunitas, layanan masyarakat, puding seledri, sedari hati

ABSTRACT

Glomus tumour, also known as angioneuromyoma, is a benign neoplasm arising from glomus bodies, a structure that functioning in thermoregulation. This rare benign tumour has been reported at a number of sites in the body, most commonly seen in the subungual areas of the digits of the hand. Only a few have been reported on the foot region, especially the great toes. Classical triad of spontaneous pain, pressure tenderness, and cold hypersensitivity is commonly found as the clinical presentation of this kind of tumour. A 33-year-old woman presented to our hospital with complaints of chronic pain and cold temperature sensitivity, in her right great toe with difficulty in wearing shoes for the last 9 years. X-rays and laboratory investigations were normal. Further investigation with magnetic resonance imaging was done and revealed regular bordered soft tissue lesion with hypo-intensity in T1 and hyperintensity in T2. Lesion removal was done by excision and a histopathological report confirmed the diagnosis of a glomus tumour of the great toe. Follow up on the outpatient clinic revealed that the patient has been pain free for 5 weeks after surgery. Glomus tumour as the possible cause of chronic toe pain is often delayed and misdiagnosed as paronychia because of its rare clinical appearances, especially on great toe. The purpose of this case report is to increase awareness of the physicians and surgeon about this tumour as the possible diagnosis.

Keywords *: community service, community midwifery, sedari hati, hypertension, celery pudding*

INTRODUCTION

Glomus tumour is a benign neoplasm arising from hyperplasia of the neuromyoarterial apparatus called glomus bodies, a contractile neuromyoarterial receptor that regulates peripheral blood flow and temperature. Glomus bodies are arteriovenous shunts surrounded by glomus cells, which contract via sympathetic activity to regulate blood flow to the extremities. These structures are highly specialized and play a vital role in thermoregulation, especially in areas exposed to environmental temperature changes, such as the fingertips and toes. Structurally, glomus bodies are composed of an afferent arteriole, an anastomotic vessel known as the Sucquet-Hoyer canal, and an efferent venule, all enveloped by layers of modified smooth muscle cells glomus cells that respond rapidly to autonomic stimuli. Glomus tumours typically arise from the modified smooth muscle cells of the glomus body and are characterized histologically by nests or sheets of uniform round cells with eosinophilic cytoplasm and centrally located nuclei. Although benign in nature, these tumours can cause significant symptoms due to their location, particularly when situated near nerves or within confined anatomical spaces like the nail bed. Patients often present with a classic triad of localized tenderness, severe paroxysmal pain, and cold sensitivity. These symptoms are often disproportionate to the small size of the lesion, making clinical detection challenging without imaging.

Despite their benign histology, glomus tumours can have a profound impact on quality of life due to chronic pain and hypersensitivity. Awareness and accurate recognition of this condition are essential for timely diagnosis and definitive surgical excision, which remains the gold standard of treatment with excellent outcomes. They are in the dermis but are more densely distributed in apical skin areas, primarily the fingertips, followed by the base of the foot and other parts of the body. Due to their high concentration in the fingers, especially under the nail bed, glomus tumours most commonly present in the subungual region and are characterized by severe localized pain, cold sensitivity, and point tenderness (Trehan et al., 2015; Valero et al., 2016). Glomus tumour has been reported at various body sites, but is most found in extremities, with up to 75% presenting in the hand, and 90% of the hand lesions located in the subungual regions of the digits. An uncommon extradigital variety is also well known. However, among the digital variety, glomus tumours have very rarely been reported in the foot regions. This rarity often leads to delayed diagnosis or misdiagnosis, as the clinical presentation in the foot may mimic other soft tissue conditions such as neuromas, ganglion cysts, or plantar fasciitis (Valero et al., 2016).

Foot glomus tumours are relatively rare due to the lower concentration of glomus bodies in the foot, even in the subungual region of the toes. Due to the low incidence and variable clinical presentations, diagnosis is often delayed. Patients may suffer from chronic localized pain, hypersensitivity to cold, and pinpoint tenderness for several years before a definitive diagnosis is made. In the foot, the clinical presentation can be easily mistaken for more common conditions such as Morton’s neuroma, plantar fasciitis, flexor hallucis longus tendon injury, plexiform neurofibroma, or even an ingrown toenail, leading to misdiagnosis and inappropriate treatment. Because of these overlapping symptoms, it is crucial for clinicians to maintain a high index of suspicion in patients with chronic, unexplained foot pain, especially if localized and resistant to standard therapies. Advanced imaging modalities, such as high-resolution ultrasound and magnetic resonance imaging (MRI), are highly beneficial in identifying glomus tumours. MRI can reveal small, well-defined lesions with characteristic signal intensity that aid in diagnosis.

Histologically, glomus tumours in the foot display similar features to those in more common locations: nests of uniform round glomus cells, a rich vascular network, and a surrounding fibrous capsule. Surgical excision remains the treatment of choice and offers

excellent prognosis with minimal recurrence if completely removed. Awareness and early detection are essential, as prompt surgical management often leads to immediate symptom relief and restores normal function, significantly enhancing patient quality of life. Despite their rarity, foot glomus tumours should be considered in the differential diagnosis of persistent digital or subungual pain (Trehan et al., 2015; Verma et al., 2021).

METHODE

This report describes the case of a 33-year-old woman with complaints of chronic pain and cold sensitivity in the right big toe for 9 years. The patient had consulted several doctors previously and was diagnosed with paronychia, but there was no improvement. Physical examination revealed swelling of the right big toe with severe tenderness on the lateral side of the nail. Radiographic (x-ray) examination showed no bone or soft tissue abnormalities & MRI examination showed a soft tissue lesion with regular borders, hypointensity on T1, and hyperintensity on T2, suggesting a possible glomus tumor. Surgical procedure with excision The lesion was removed by excision after partial removal of the nail plate. The lesion was 5×4×3 mm in size, pale red in color, and showed no signs of invasion into surrounding tissues. The removed tissue sample was sent for histopathologic examination.

RESULT

The glomus tumour was first reported by Wood in 1812 as a ‘subcutaneous tubercle’, but a complete description was mentioned by Masson in 1924 (Valero et al., 2016). Glomus tumour is a benign neoplasm originating from a neuromyoarterial apparatus called glomus body. Glomus bodies are arteriovenous shunts that surrounded by glomus cells, present in the Sucquet-Hoyer canal, which contracts via sympathetic activity, thereby regulating blood flow to the extremities (Mohindra et al., 2016; Polo et al., 2012; Verma et al., 2021). Glomus bodies functioning in thermoregulation, decreasing blood flow when the body is exposed to cold and increasing it when it is hot. Glomus bodies are located in the dermis throughout the body but more densely concentrated in apical skin areas, primarily the fingertips of hand, followed by the base of foot and the rest of the body. The most common site of occurrence of this tumour is the subungual area of the fingers, and it may constitute almost 2% - 5% of all hand tumours with 75–90% of these in subungual locations (Trehan et al., 2015). Only few studies have been reported this tumour in toes (Heys et al., 1992; Polo et al., 2012).

Glomus tumours is more common in females in the age group of 30 to 50 years than in males. Glomus tumours have been categorized into solitary and multiple types. Most 90% have been solitary, with up to 10% multifocal involvement can occur . Solitary glomus tumours will be encapsulated, located in subungual locations, and contain numerous small vascular laminae. In contrast, the multiple type tumours will be unencapsulated, will rarely be subungual, will have larger shaped vascular spaces, and will often be asymptomatic. Intraosseous glomus of the toe nail has also been reported, where the glomus bodies are located in the medullary canal of the distal phalanges. Familial occurrence of the tumour has also been reported (Folpe et al., 2001; Kleihues & Cavenee, 2000).

Glomus tumours are composed of glomus cells, small vessels, and smooth muscle cells in various combinations. Microscopically, the lesion comprises uniform cells, epithelial in nature distributed outside an abundant branching vasculature. Although 4 types are reported (angiomatous, paucivascular, neuromatous, and mucoid hyaline) an exact categorization is often not possible due to frequent mixture of various types. However, the angiomatous component is the most dominant. The capsule is a pseudo capsule formed by condensation of surrounding tissue and may show infiltration in 1% to 2% of cases, but generally without any

evidence of malignant change. Malignant degeneration is reported less than 1% for lesions that are >2 cm or located deep within the dermis (Trehan et al., 2015; Verma et al., 2021).

DISCUSSION

The tumour has both myelinated and nonmyelinated nerve fibres entering the capsule, shown to be capable of releasing neurotransmitters such as substance P. Distortion of the capsule because of changing vascularity generates a bursting pain, which at times can be severely disabling and the morbidity caused by this lesion is debilitating (Heys et al., 1992; Polo et al., 2012). About 1% of all glomus tumours are malignant with histopathological features include large size (2 cm) and deep location, or moderate-to-high nuclear grade and increased mitotic rate, or the presence of atypical mitotic figures. If the histological criteria for malignancy are met, then the lesions are associated with more than a 25% risk of metastasis. However, tumours with some but not all of these features (high mitotic activity and superficial location, large size only, or deep location only) are labelled as glomus tumours with uncertain malignant potential, or symplastic glomus tumors (high nuclear grade only) (Folpe et al., 2001).

The key to the diagnosis of glomus tumour generally presents with a classical triad of intense paroxysmal pain, pinpoint tenderness, and hypersensitivity to cold. Pain is the most common symptom and is generally sharp and bursting in nature and may radiate to the base of the toe. A bluish discoloration below the nail plate may be present or a palpable mass that deforms the nail plate (Carroll & Berman, 1972). However, not all glomus tumours will present with this classic triad of symptoms. Van Geertruyden et al. 1996 have reported that pain, point tenderness, and cold sensitivity will occur at a rate of 80%, 100%, and 63%. A history of preceding trauma may be existed, but connection with previous injury has not been established yet, although some authors have hypothesized that injury causes weakness of the glomus and a reactive hypertrophy (Samaniego et al., 2009; Van Geertruyden et al., 1996).

Several test can be useful in clinical examinations to diagnose glomus tumours. The Love test is performed by probing with a needle or pointed instrument triggers pain in the affected area but not the area immediately adjacent to it. The test has a sensitivity of 100% but a specificity of 0% (Mundada et al., 2019). The Hildreth sign consists of inducting ischemia by applying a tourniquet above the systolic pressure; if the pain disappears, it is probably a sign of glomus tumour. This highlights the vascular nature of the lesion. It has a sensitivity of between 77.4% and 92% and a specificity between 91% and 100% (Samaniego et al., 2009). Cold-sensitivity test has a sensitivity and specificity of 100%, is carried out by applying of cold water or ethanol to the affected area will also reproduce the symptoms. Transillumination test is performed in a darkened room by passing light through the finger pad. An opaque red image is observed in the region of the tumour that allows estimation of its size. The test has a sensitivity up to 38% and a specificity of 90% .

The radiolographic investigations are pathognomonic if there is osseous erosion caused by the tumour lesion. Plain radiography features include bone erosion, intra-osseous lytic lesion, and even only widening of the distance between the nail and bone. Plain x-ray radiographs have limited utility due to only 36% of patients have associated bone erosion (Dahlin et al., 2005; Mundada et al., 2019). Ultrasonography of subungual glomus tumours are hard to evaluate as compared to those in periungual location. The lesion appears as a non-specific hypoechoic solid mass, often associated with erosion of underlying bone. Doppler imaging shows high-velocity flow in the most, but not in all, of the cases (Matloub et al., 1992).Magnetic resonance imaging (MRI) is useful to asses tumour location at pre-surgical mapping for treatment planning, clinical suspicion associated with bone erosion, and signal characteristics. A dark outlined image is seen on T1 sections and a bright outlined mass is noticed in T2 sections. On dynamic MR contrast angiogram, the lesion shows intense

enhancement in the arterial phase with tumour blush. Al-Qattan et al reported that MRI sensitivity was 90%, specificity was 50%, positive predictive value was 97%, and negative predictive value was 20% (Al-Qattan et al., 2005).

A conclusive diagnosis requires a histopathological examination. The findings in this case are similar to research conducted by Patel et al. (2024) a homogeneous epithelium with an abundance of capillaries is the microscopically visible feature of benign glomus tumors. Glomus cells are small, homogenous cells with spherical, eosinophilic cytoplasm, and monomorphic nuclei. Benign and malignant glomus cells often have a similar appearance. Malignant glomus tumors in the foot are very rare, but histopathological evaluation must still be carried out routinely (Gombos & Zhang, 2008; Patel et al., 2024). The diagnosis is essentially clinical, usually confirmed on histopathology with surgical excision. A clear history, meticulous examination, and modern investigations should be taken to diagnose the tumour early (Kreel et al., 1986). However, because subungual glomus tumors are located beneath the nail plate, they cannot be detected through visual inspection or palpation. This can lead to significant delays in diagnosis or even misdiagnosis, with patients sometimes being incorrectly labeled as malingerers or neurotics. It is common for patients to be initially diagnosed with chronic paronychia and treated for this condition over extended periods (Chen et al., 2020; Kreel et al., 1986).

Paronychia refers to the inflammation of the tissue surrounding the nail and can be classified as acute (lasting less than 6 weeks) or chronic (lasting more than 6 weeks). Acute paronychia typically results from a break in the protective barrier of the nail folds, allowing pathogens to cause inflammation and infection. Common causes of nail fold disruption include trauma, artificial nails, manipulation of hangnails, onychocryptosis (ingrown toenails), and onychophagia (nail biting). The causes of acute paronychia bacteria include Fusobacterium, Pseudomonas, Bacteroides, Eikenella, Enterococcus, Klebsiella, Proteus, Streptococcus, and Staphylococcus. Acute paronychia caused by Pseudomonas can cause a greenish discoloration at the base of the nail. On the other hand, the tumor glamour appears erythematous which is suggestive of vascular problems. Diseases such as pemphigus vulgaris, pustular psoriasis, and reactive arthritis can also cause acute paronychia.

Various drugs also contribute to the formation of acute paronychia such as (EGFR) inhibitors, cytotoxic chemotherapeutic agents (taxanes, doxorubicin, methotrexate, capecitabine), antiretroviral agents, and systemic retinoids. Acute paronychia appears within 1-3 months after the first administration of these drugs. Patients with acute paronychia typically present 2 to 5 days after the initial cause, showing symptoms such as acute onset of redness, swelling, pain, and tenderness along the lateral and/or proximal nail folds, usually affecting a single digit. The presence of psoriasis, Reiter's syndrome, and pemphigus involving multiple digits with characteristic nail changes may be a useful marker of acute paronychia. The diagnosis of acute paronychia is often made clinically. Cultures of the fluid obtained are always nondiagnostic and do not affect management. Treatment depends on the severity of inflammation and the presence of an abscess, ranging from topical or oral antibiotics to abscess drainage. If a subungual glomus tumor is misdiagnosed as acute paronychia, standard treatments will not alleviate the pain, which will persist (Alessandrini et al., 2017; Leggit, 2017; Relhan & Bansal, 2022).

Chronic paronychia, on the other hand, is characterized by inflammation lasting more than 6 weeks. It differs from acute paronychia because it results from irritant dermatitis rather than an infection. Common irritants include acids, alkalis, and other chemicals frequently encountered by housekeepers, dishwashers, bartenders, laundry workers, florists, bakers, and swimmers. Disruption of the protective nail barrier followed by repeated exposure to irritants can lead to chronic inflammation. Immunosuppressive conditions such as diabetes mellitus, HIV can cause secondary colonization. It is also susceptible to less common causes such as

bacterial, fungal (candida albicans), viral, and secondary syphilis. Diagnosis is based on a symptom duration of at least six weeks, a history of exposure to irritants, and clinical signs consistent with nail dystrophy, often involving multiple digits. Chronic paronychia can cause erythema, pain, swelling, and pus discharge that can involve more than one fingernail. In addition, ascending matrix damage can lead to discoloration, ridging, Beau's lines, and onychomadesis. If only one digit is affected, malignancy, such as squamous cell carcinoma, should be considered. Treatment involves removing the source of irritation, controlling inflammation, and restoring the protective barrier. Topical anti-inflammatory agents, steroids, or calcineurin inhibitors are commonly used. Neoplasms and malignancies of the nail unit may present as paronychia, and a lack of response to conventional treatments is an omnious sign (Leggit, 2017; Relhan & Bansal, 2022).

Other differential diagnoses include ganglion cyst, hidradenomas, neuroma, angioma, myxoid cyst, melanoma, chronic paronychia, gout arthritis, granuloma, subungual melanoblastoma, subungual hematoma. Other less possible diagnosis is neuron, plexiform neurofibroma, papilloma, and fibromas and Koenen's tumours (Kreel et al., 1986). Misdiagnosed of glomus tumor as paronychia lead to ineffective and unsuccessful surgical treatment as it only removing small portion of the side of the nail and destroying the nail bed beneath. To achieve complete resolution of the symptoms, surgical treatment of glomus tumour requiring surgeon to completely excise the mass. The mass may be accessed via the nail plate avulsion or a periungual approach depending on the location. A trans ungual extraction (through a window) has been described, but is generally difficult due to dystrophic nail. Post operative recovery takes about 2 to 4 weeks, but at times, it may take a couple of months for the patient to be completely pain free.

Glomus tumour recurrence is very uncommon. Causes of recurrence are incomplete resection, missed diagnosis of coexisting multiple glomus tumour and development of new glomus tumour at the excision site. The development of a second tumour contributes for late recurrence. The prognosis for the hand lesions is low but significant recurrence rate of 11.9% as reported by Conant et al. (1971) and 17% by Lin et al. (2010), but no author has been reported recurrence of the toe regions (Conant & Wiesenfeld, 1971; Lin et al., 2010).

CONCLUSION

In conclusion, to successfully diagnose foot glomus tumours needs vast clinical knowledge, given its rarity and low incidence and wide differential diagnosis. Glomus tumour should always be kept in mind as a rare but possible differential, particularly if a young or middle-aged woman presents with a painful lesion of the toes that didin't resolves with conventional treatment of paronychia. History-taking related to risk factors, onset, progression, and physical examination is essential to differentiate glomous tumors and other diagnoses including paronychia. Late diagnosis is commonly seen and often the patient suffers great morbidity for years and having knowledge of this kind of tumour can help to reduce it. Clinical examination manoeuvres may be helpful in diagnosis. MRI is recommended to help confirm the diagnosis, also as to rule out multiple glomus tumours within the same region. Surgical tumour excision could result in a complete symptom relief and cure.

Through this case report, we want to raise awareness of glomus tumours and emphasize the need for an early diagnosis and treatment, to reduce the great morbidity suffered by patients. This case showed that glomus tumours are small but very painful tumours, challenging to diagnose and often the patient is misdiagnosed between different physicians. However, with proper history, examination, and investigations, this tumour can be diagnosed early and successfully managed.

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