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Extradigit Glomus Tumor Causing Abdominal Pain
-A Case Report-

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Glomus tumors are small vascular tumors that are usually benign and rarely occur. They originate from glomus bodies and present in the reticular dermis. They are clinically distinguished by their small size and their ability to cause extreme pain. Most of these tumors are subungually located. However, atypical locations of the tumors sometimes cause misdiagnosis, particularly when the lesion is rarely reported. Therefore, we report a case of glomus tumor which presented with chronic abdominal pain, found in the abdominal wall that has never been reported before. (Korean J Pain 2012; 25: 108-111)

Key Words:
abdominal pain, abdominal wall, glomus tumor.

Chronic abdominal pain has multifactorial etiology and should be carefully examined by a pain physician especially when the pain source has nothing to do with medical condition. Glomus tumors are small vascular tumors which are benign, but rarely occur [1]. Approximately 80% of the tumors are located in the upper extremities, and more than 75% of these are located subungually [2]. Many extradigit locating glomus tumors have been reported in various reports [3,4]. Although pain is the major complaint of the patient, glomus tumors are often overseen by pain physicians. Here, we report a case of glomus tumor which presented with chronic abdominal pain, located in the abdominal wall.

CASE REPORT

A 71-year-old man visited our pain clinic presenting with a 3-year history of abdominal pain. The nature of pain was constant, superficial throbbing and lancinating and sometimes diffusely radiated with visual analogue scale (VAS) score 70-80 of 100. The location of the tumor was on the right lower quadrant of abdomen. The patient complained of tenderness on the area but the pain developed even spontaneously. It was more severe at night and early morning, thus resulting in sleep disturbance. There was no traumatic history including medical operation on the pain area. The patient had no specific medical history

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except pain killers due to the pain described above. Previous treatment included non-steroidal anti-inflammatory drugs (NSAIDs), opioids, local heating, bracing, epidural block, peripheral block and local anesthetic infiltrations, and had no effect. Imaging studies including X-ray, abdominal ultrasonography and magnetic resonance imaging (MRI) showed non-specific findings. He was taking anticonvulsant medications under the diagnosis of post herpetic neuralgia (PHN) at the hospital he had previously visited. Neurological examination and routine laboratory tests showed normal findings. No gastroenterological problem could be found as the cause of chronic abdominal pain after consultation to the department of internal medicine. Physical examination was characterized by exquisite pressure tenderness over the painful area on the right lower quadrant. A careful examination of ultrasonography over the painful area showed a 4.7 × 3.6 mm^2 small nodular lesion with increased focal blood flow, and a well-circumferenced hypoechoic character. Suspicions of infected sebaceous cyst or epidermal cyst were made (Fig. 1). We did not think of this cyst as the origin of the patient's pain. Increasing anti-convulsant medication did not show any effect. NSAIDs showed some effectiveness but the patient failed to be free of pain. Local anesthetic injections around the cyst decreased his pain to some degrees (VAS score 60 of 100). Nevertheless, injections into the cyst developed more severe pain up to VAS score 100. In spite of repetitive treatment, his symptoms did not improve. With the possibility that this cyst could be the source of his pain, we transferred the patient to the department of surgery for a detailed evaluation. A general surgeon carefully followed up for about 5 months with conservative treatment including steroid and painkillers under the suspicion of thrombophlebitis. However, he did not seem to get any better,

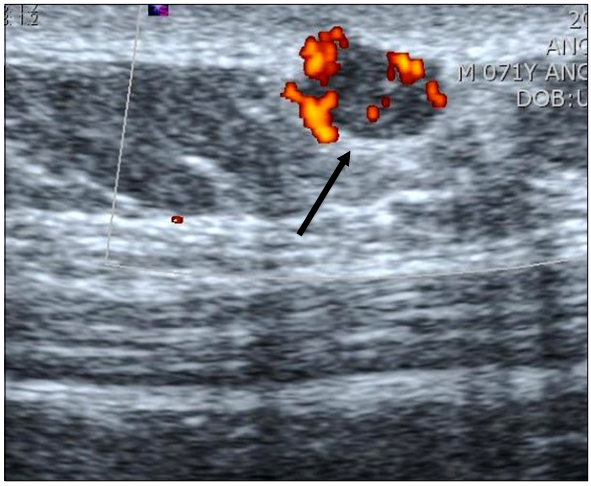

Fig. 1. Ultrasonography imaging. Revealed a 4.7 × 3.6 mm^2 sized nodule with increasing focal blood flow, which had a well-circumferenced hypoechoic character suspicious of infected sebaceous cyst or epidermal cyst.

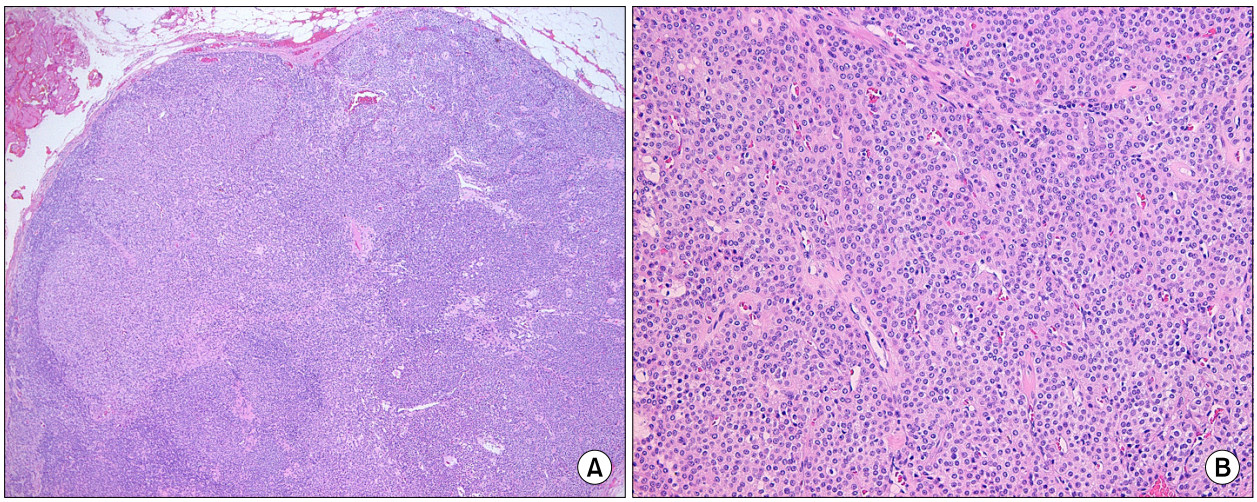

Fig. 2. (A) The lesion consists of polygonal cells that are well encapsulated with areas of cystic changes. The glomus tumor was a relatively well-demarcated lesion arising from the dermis, anddilated veins were not shown (stain; hematoxylin and eosin; original magnification, ×40). (B) The cells are monotonous and cuboidal with rounded nuclei and eosinophilic cytoplasmand the nuclei were round to oval and delicate with chromatic margins (stain; hematoxylin and eosin; original magnification, ×200).

maintaining a VAS score of 80 out of 100. Finally the surgeon decided to proceed with diagnostic excision. It was confirmed as a 14 × 10 × 7 mm^3 glomus tumor by histologic examination (Fig. 2). The excision was completely curative, and the patient was free of pain, with a VAS score of 0 out of 100 without any medication.

DISCUSSION

Glomus tumors were first described clinically by Wood in 1812 as "painful subcutaneous tubercles" [3]. They are rare benign tumors originating from the neuromyoarterial glomus bodies of Masson. Although they can be found anywhere in the body, about 75% of them are found in the hands. The most common site is the distal phalanx, especially the ungual bed. Thus glomus tumors were usually recognized as a cause of digit pain, especially in the subungual region. Tumors may not be palpable because they are very small, often 3 to 4 mm in average size and sometimes deeply located in the tissue. Middle-aged women have been known to be more frequently affected than men. Surgical removal is curative and recurrence is rare [5,6]. They have been thought to be less common in extradigital locations. Therefore, diagnosis of extradigit glomus tumors is commonly delayed or even missed. In a previous report [7], the average duration of symptoms was up to 10 years. During this time, consultations with an average of 2.5 physicians were routinely done until an exact diagnosis was made. The prognosis was extremely good if they were properly treated.

The classic diagnostic triads are known as spontaneous pain, pressure tenderness, and cold hypersensitivity [8]. When the tumor can be detected by physical examination, there is a reddish or bluish discoloration of the skin, or contact produces a sharp pain that radiates diffusely. Pain according to temperature variation also can be a clue to diagnosis. The physician primarily depend on above symptoms in the diagnosis of glomus tumors. Two clinical diagnostic tests are known to be helpful. One of them is "Ischemia test" [9]. The diagnosis of glomus tumors is strengthened if inflation of a blood pressure cuff to above systolic pressure completely reduces pain after several seconds. Unfortunately, this test can be applied only in the upper extremity region. Another test is Love's test that consists of severe local pressure tenderness elicited when applying pressure with a blunt point to the patient's painful area [10]. Clinical differential diagnosis includes neuroma, Raynaud's phenomenon, infection, gout, peripheral neuropathy and radiculopathy [11]. On the histological aspect, hemangiopericytoma, hemangioma, angiomyoma, neurilemoma, and many other hamartomas are under differential diagnosis [12]. In our case, the reason for conservative therapy by the general surgeon was aspiration cytology results only showed a blood clot and thrombophlebitis also considered to be one of the causes of pain, including above differential diagnostic diseases. MRI has known to be the most sensitive imaging modality for diagnosis of glomus tumors but specificity is limited to about 50% [3]. Radiography, angiography, thermography, ultrasonography and scintigraphy have been used to diagnose glomus tumors but with limited diagnostic value [3].

In our case, due to the chronic abdominal pain, differential diagnosis mentioned above such as gastrointestinal disorders were also ruled out after imaging studies and internal medicine consultation. Also, the patient did not satisfy the diagnostic triad in that and no discoloration could be found. However, the Love's test was positive at physical examination. To evaluate the characteristics of the observed cystic lesion, we injected local anesthetics around and into the cystic lesion. Injection around the cystic lesion brought relief of the pain. However, injection into the lesion provoked more severe pain with strong resistance, respectively. Histological characteristics of glomus tumors differentiate them from other tumors. Glomus tumors cells have properties that are similar to smooth muscle cells, accounting for their contractility which is very sensitive to pain [13]. Thus injection into the tumor will produce severe pain. Addition to symptom triads, Love's test and "injection local anesthetics", the final diagnosis was made with excisional biopsy.

Early diagnosis of glomus tumors, especially in rare locations, would help patients from suffering pain for a long time and conducting unnecessary procedures. Pain physicians should include glomus tumors in the differential diagnosis of any localized tenderness associated with extreme neuropathic pain. Excisional biopsy should be done to make a confirmation diagnosis when a suspicious lesion is found during examinations. Furthermore, excision will be a curative management for the lesion. This case raises the awareness of atypical causes of regional pain syndromes related to glomus tumors.

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