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Extra digital glomus tumor: A very rare cause of chronic abdominal wall pain

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ABSTRACT

INTRODUCTION: Glomus tumors are very rare benign vascular tumors, constituting less than 2% of soft tissue tumors. These tumors originate from the glomus body. 75% of these tumors occur in hand however rarely can be found in any body part.

PRESENTATION OF CASE: We here report a case of glomus tumor who presented with abdominal pain (constant and throbbing nature) and small swelling in the left hypochondrium. Pain was mostly spontaneous without any obvious cause, aggravated by cold and palpation. Ultrasonography parietal wall showed 27 × 22 × 21 mm hypochoic lesion in the parietal wall with increased focal vascularity. Histopathological examination confirmed the diagnosis of glomus tumor.

DISCUSSION: These are rare benign vascular tumors arising from the glomus bodies found anywhere in body. However 75% are found in hand mostly subungual region. Glomus tumor may show unusual clinical picture such as extra digital location, large size, deep soft tissue, visceral location, multi-centric or infiltrative growth pattern. These tumor commonly presents with a diagnostic triad of spontaneous pain, hypersensitivity to drop in temperature and pressure tenderness. Clinical diagnostic tests aide in diagnosis, including Love's test, Hildreth's test, Transillumination and the cold test. The clinical differential diagnosis of glomus tumor includes Raynaud's phenomenon, neuroma, gout, infection, peripheral neuropathy and radiculopathy.

CONCLUSION: Extra digital glomus tumor occur in any part of the body and should be put in differential diagnosis of abdominal pain when no obvious cause of pain is found. Surgical excision is the curative treatment of choice with rare recurrence.

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1. Introduction

The glomus body is an arteriovenous shunt responsible for thermoregulation. Glomus tumors (GT) are very rare benign tumors, constituting less than 2% of soft tissue tumors [1]. Almost 80% of the GT arise from the upper extremities and 75% are located in the subungual region [2]. Many extra digital locations have been reported in many case reports [3]. Pain is the most common presenting symptom of these patients. This work has been reported in line with the SCARE criteria [4].

2. Case report

2.1. Patient information

A 70 year male patient presented with chief complaints of abdominal pain from last 2 years. Pain was constant and throbbing in nature, sometimes diffusely radiating. Pain was more severe during the night time and early morning, thus disturbing in sleep. Pain was also aggravated when patient had bath in cold water. Pain was mostly spontaneous without any obvious cause. There was no significant medical, psychosocial or family history. There was no history of previous trauma or abdominal surgery.

- Drug history, family history including any relevant genetic information, and psychosocial history (item 5d).

2.2. Clinical findings

General physical examination was grossly normal. Local examination revealed a swelling about 3.0 × 2.5 × 2.0 cm in abdominal

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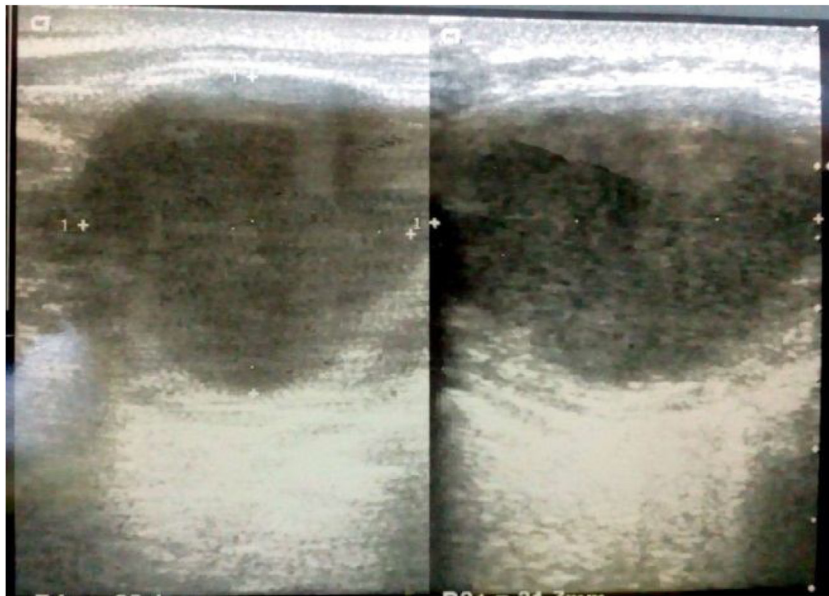


Fig. 1. Ultrasonography imaging. Revealed 27 × 22 × 21 mm hypoechoic lesion in the muscular plan of abdominal wall with increased local blood flow.

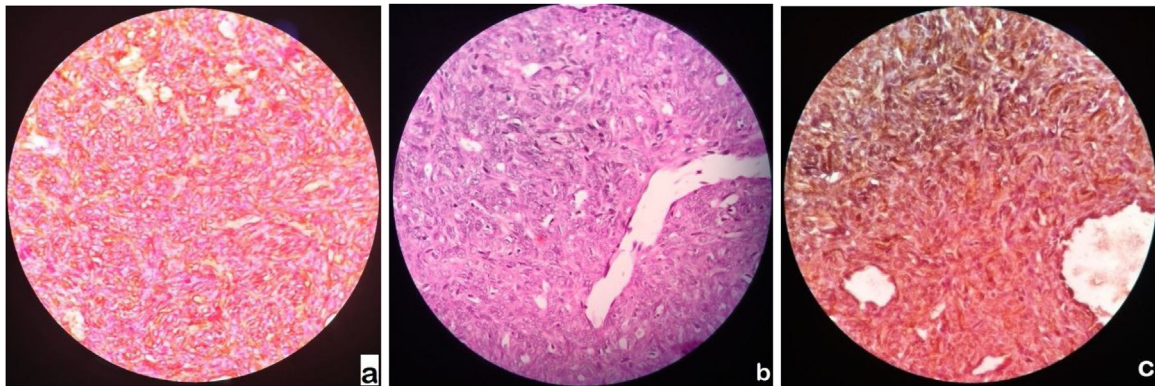


Fig. 2. Histopathological examination revealed Glomus Tumor (2a - showing staining of vascular spaces by CD34, 2b - Small uniform round cells arranged in sheets and around vascular spaces, 2c - showing Smooth Muscle Antigen (SMA) positive glomus cells).

wall, left hypochondrium without any overlying skin changes. There was no visible peristalsis or prominent veins over the swelling. Temperature over the swelling was normal. Local tenderness was present over the swelling with aggravation of pain on deep palpation. It was soft in consistency with overlying skin freely mobile. Swelling was non-pulsatile. Auscultation does not reveal any bruit. Transillumination test was negative and the Carnett's test was positive. Ultrasonography (USG) abdomen and parietal wall showed 27 × 22 × 21 mm.

2.3. Diagnostic assessment

Ultrasonography (USG) abdomen and parietal wall showed 27 × 22 × 21 mm hypoechoic lesion in the parietal wall with increased focal vascularity (Fig. 1). Routine laboratory tests were normal. No obvious gastroenterological cause of pain was found.

2.4. Therapeutic intervention and follow up

Excisional biopsy was done which revealed features of glomus tumor (Fig. 2). After complete excision of the lesion the patient became pain free. Patient is on routine follow up and is doing well.

3. Discussion

The glomus cells with smooth muscle property form the glomus body, regulating thermoregulation by controlling blood supply to the skin. Glomus tumors are rare benign vascular tumors arising from the glomus bodies in reticular dermis [3]. Although These tumors are found anywhere in body, about 75% are found in hand mostly subungual region of digits, often presenting as painful subcutaneous nodules which are purple in color. Occasionally glomus tumors may show unusual clinical picture such as extra digital location, large size, deep soft tissue, visceral location, multi-centric or infiltrative growth pattern [5]. Therefore the diagnosis is often delayed or even missed. Commonly affects middle aged females than males. Glomus tumor commonly presents with a diagnostic triad of spontaneous pain, hypersensitivity to drop in temperature and pressure tenderness [6]. Pain with drop in temperature can be a clue to the diagnosis. There are few clinical diagnostic tests which may aide in diagnosis, including Love's test, Hildreth's test, Transillumination and the cold test. Love's test pinpoints the location of tenderness. Hildreth's test shows completely reduction of pain after few seconds of inflation of blood pressure (BP) cuff above the systolic BP. Transillumination of digit and observation of an opaque reddish mass with an illumination. Cold test shows provocation of

pain on application of cold water or ethyl alcohol. MRI is the most sensitive imaging modality for glomus tumor [7]. USG with high frequency transducer is most useful when the tumor size is less than 2 mm with a positive predictive value of 100%. The clinical differential diagnosis of glomus tumor includes Raynaud's phenomenon, neuroma, gout, infection, peripheral neuropathy and radiculopathy. Histological differential diagnosis include angiomas, hemangioma, hemangiopericytoma, neurilemoma and other hamartomas.

Glomus cells histologically present with eosinophilic cytoplasm and oval nuclei after staining with haematoxylin adenosine. These tumors immunohistochemically stain positive for neuron-specific enolase, vimentin, and muscle actin and negative for S100 and cytokeratin [8]. These tumor cells are usually immunoreactive for CD34. Surgical excision is the treatment of choice with a rare recurrence [9].

Our case presented with chronic abdominal pain. The differential diagnosis was oriented around the disorders of gastrointestinal tract (GIT) and parietal wall. GIT disorders were ruled out by investigations and surgical and medical gastroenterology consultation. The diagnostic triad of GT was present with local pain aggravation on bathing with cold water, local tenderness and spontaneous pain. Love's and cold tests were positive while as Hildreth's test and Transillumination was not possible in our patient. Surgical excision completely cured our patient. Pain score after excision was 0 out of 10 on VAS. Final diagnosis was made by excisional biopsy. Histopathological examination revealed vascular tumor with round to irregular open blood vessels surrounded by round to spindle cells with moderate eosinophilic cytoplasm. Vimentin and smooth muscle actin were strongly positive, CD 34 faint positive and keratin (CK7) was negative. Surgical excision is the curative treatment of choice and the recurrence is very rare.

4. Conclusion

Extra digital glomus tumor can occur in any part of the body including abdominal wall. It should be put in the differential diagnosis of abdominal pain when there is no obvious cause found. Whenever suspicious lesion is found during examination excisional biopsy should be done, as the gold standard diagnosis for these lesions is histopathological examination and immunohistochemical markers. Surgical excision is the curative treatment of choice with rare recurrence. Our case has raised the awareness of atypical cause of regional pain syndrome related to glomus tumor.

Patient's perspective

I had pain in the left upper part of my tummy which was aggravated when even I had bath in cold water. This pain was irritating me and I was not able to do daily routine work normally. After surgery the pain in my tummy disappeared and I can go to my routine work without any pain and I can have bath in cold water as well. I am very thankful to my doctors who had removed this tumor and made my life more comfortable.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dar Parvez Mohi Ud Din: Performing the surgery, Final approval of the version to be submitted. Wani Afshan Anjum: Acquisition of data and analysis of data. Malik Liaqat Ahmad: Interpretation of data. Suhail Mushtaq: Conception and design the study, drafting the article.

Registration of research studies

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