Annals of Case Reports
Wirth T, et al. Ann Case Rep: 9: 101592
www.doi.org/10.29011/2574-7754.101592
www.gavinpublishers.com

Case Report

Glomus Tumor Inside Xyphoid Appendices: A Case Report

Theo Wirth[1,2*], Pierre Mathieu Bonnet[3], Christophe Chagnaud[4], Nathalie Balandraud[1,2]

[1]Rheumatology Department, Sainte Marguerite Hospital, Aix-Marseille University, APHM, Marseille, France

[2]Autoimmune Arthritis Laboratory, INSERM UMRs1097, Aix Marseille University, Marseille, France

[3]Department of Thoracic Surgery, European Hospital, Marseille, France

[4]Department of Radiology, Conception Hospital, Aix-Marseille Univ, Marseille, France

[*]**Corresponding author:** Theo Wirth, Rheumatology Department, Sainte Marguerite Hospital, Aix-Marseille University, APHM, Marseille, France

Citation: Wirth T, Mathieu Bonnet P, Chagnaud C, Balandraud N (2024) Glomus Tumor Inside Xyphoid Appendices: A Case Report. Ann Case Report 9: 1592. DOI: 10.29011/2574-7754.101592

Received: 10 January 2024; **Accepted:** 16 January 2024; **Published:** 19 January 2024

Abstract

This case report outlines a distinctive presentation of a glomus tumor in the sternocostal joint of a 35-year-old woman with cutaneous psoriasis. Despite the initial suspicion of Tietze syndrome, clinical and imaging assessments, particularly MRI, led to the diagnosis. The patient underwent successful surgical resection, resulting in complete recovery. Anatomopathological analyses confirmed the diagnosis of a glomus tumor. This case emphasizes the importance of considering glomus tumors in atypical locations when chest pain persists, highlighting the role of advanced imaging for accurate diagnosis and subsequent curative management.

Keywords: Glomus Tumor; Xiphoid Appendice; Costal Cartilage; Nonsteroidal Anti-Inflammatory Drugs (NSAIDs)

Case Presentation

We present here the case of a 35-year-old woman with a history of cutaneous psoriasis and an otherwise unremarkable medical background. The patient reported anterior chest pain persisting for 14 months, initially triggered after a three-week bout of coughing, and exacerbated, notably, following an osteopathic session. The pain progressed to a severe level, imposing functional limitations and hindering her ability to work. Despite the distressing symptoms, there were no accompanying signs of fever, weight loss, or asthenia. Traditional pain relievers such as nonsteroidal anti-inflammatory drugs (NSAIDs) and paracetamol offered no respite. The patient found physical reeducation sessions intolerable, and symptoms were exacerbated by upper limb movement or direct contact but alleviated with rest. Physical examination revealed tenderness in the right para-xyphoid area and identified a "trigger" point in the intercostal joint, with an otherwise unremarkable general examination and no lymphadenopathy.

Clinical observations led to a provisional diagnosis of Tietze syndrome. Initial treatment with NSAIDs and paracetamol, however, proved ineffective, prompting further investigations. Ultrasound echography revealed a 2 x 6.5 mm tissue formation with central hypo-echogenicity behind the last right chondro-costal joint, displaying concomitant degenerative joint changes. Mammography using MRI exhibited an intra-articular chondro-costal lesion, while X-ray mammography and Tm99 scintigraphy yielded no abnormalities. Computed tomography (CT) scan results disclosed a 6 mm x 3 mm ovoid lesion with low density and

minimal contrast enhancement post-contrast injection. Subsequent MRI focusing on the sternal bone displayed the lesion with iso-signal intensity on T1-weighted images, hyper-signal on STIR images, and enhancement after gadolinium injection (Figure 1).

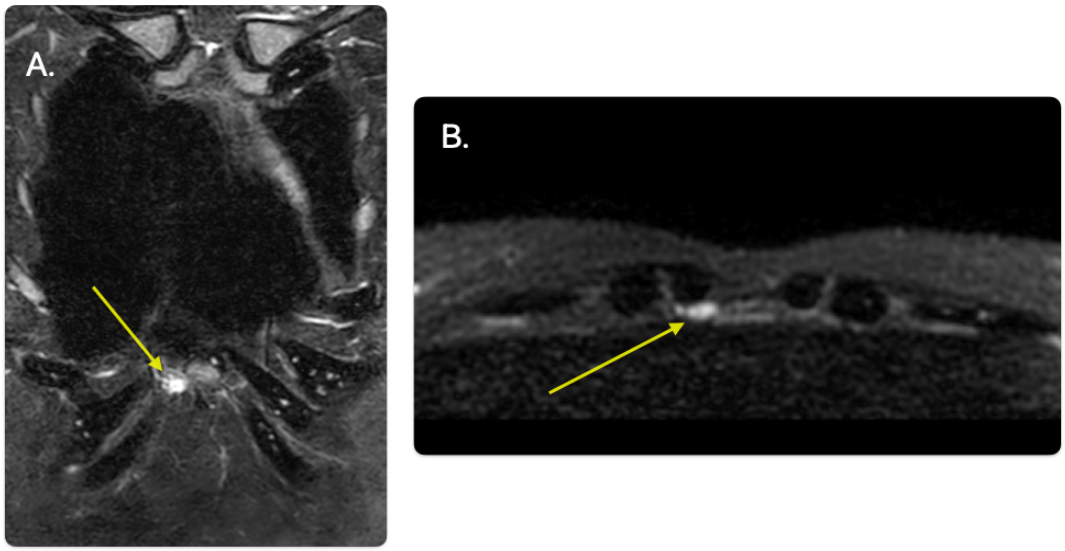

Figure 1: Tissue-like image measuring 11x8 mm in the coronal plane with a 3 mm anteroposterior diameter, localized over the manubriosternal joint, exhibiting STIR hypersignal on: A. Coronary section B. Axial section.

Laboratory analyses identified the presence of antinuclear autoantibodies (titer: 1/80) and asymptomatic hypocalcemia (2.17 mmol/L), with no other autoantibodies or elevated inflammatory markers detected. Following a multidisciplinary consultation, the patient underwent surgical intervention involving a monobloc resection of the xyphoid and the medial border of the right costal cartilage. The procedure was complicated by a pleural breach, necessitating a 24-hour pleural drain placement. Hospitalization lasted 48 hours without any complications during the follow-up period. Histological examination disclosed a 4 mm intraosseous angiomatoid hyperplasia, with cells exhibiting immunoreactivity to Smooth Muscle Actin (SMA). Medullary hyperplasia was also noted without signs of malignancy. The combined findings from MRI patterns and histological examination led to a final diagnosis of a glomus tumor. Three months post-surgery, the patient achieved complete recovery.

Discussion

Glomus tumors, constituting less than 2% of soft tissue tumors, are rare benign entities originating from glomus bodies. These bodies are small thermoregulatory organs surrounded by nerve elements and cells that resemble modified smooth muscle cells [1]. Typically located in the dermis and pre-coccygeal soft tissue, glomus tumors are most commonly reported in the subungual area, as well as the palm, wrist, forearm, and foot, where normal glomus bodies are abundant [1].

The existence of normal glomus bodies was initially described by Hoover in 1877, and the first clinical depiction of glomus tumors was provided by Masson in 1924 [2]. The precise etiology of glomus tumors remains elusive, although some authors have suggested a structural weakness in glomus bodies, leading to reactive hypertrophy following trauma. Genetic involvement in the pathogenesis of glomus tumors has also been proposed by several authors [3].

Glomus tumors are predominantly solitary (more than 90% of cases). Multiple involvements, referred to as glomangiomatosis, are more commonly observed in pediatric populations and are associated with chromosome 1 mutations or neurofibromatosis 1 [4]. Typically, patients present with a small nodule displaying varying hues from bluish to red, dependent on the depth of the lesion. Glomus tumors induce pain, intensified by pressure and exposure to cold. Two clinical tests aid in diagnosis: the “love” test, involving gentle palpation of the lesion to elicit pain, and the “Heldrith” test, demonstrating a decrease in pain when the affected limb segment is elevated and a resurgence of pain upon the return

of blood flow. These clinical findings are indicative of the pre-histologic diagnosis in typical locations (such as subungual and deep dermal sites). However, painless and larger lesions have also been documented, and imaging techniques can assist in diagnosis [5].

Radiographic investigations may uncover a cortical defect [6], yet, in the majority of cases, they fall short of providing a definitive diagnosis [7]. Ultrasound reveals hypoechoic lesions with posterior acoustic enhancement, and erosion or rupture of the cortical bone may be observed [8]. Magnetic Resonance Imaging (MRI) proves particularly valuable in atypical locations, showcasing characteristic features such as small lesions (< 10mm) with high signal intensity on STIR and T2-weighted sequences, low signal on T1-weighted images, and avid enhancement following intravenous gadolinium administration [9]. MRI stands out as the most sensitive and specific imaging test, boasting a specificity of 90%, sensitivity of 50%, positive predictive value of 97%, and negative predictive value of 20% [4]. Immunohistochemical analysis of the lesion enables the diagnosis of a glomus tumor. Nearly all glomus tumors are positive for alpha-smooth muscle actin (a-SMA) and muscle-specific actin (MSA), while being negative for CD31, cytokeratins, and S100 [7].

Glomus tumors typically affect women aged 30-50 in typical locations. However, in extra-digital locations, the male sex exhibits a higher incidence [10]. Lesions in atypical locations are larger and have a greater potential for malignancy [7]. Diagnostic delay is not uncommon in these atypical locations.

While previous cases have described glomus tumors involving the chest wall, this is the first reported case within the costal joint. Other instances of thoracic involvement also noted the presence of coughing, which may be a contributing factor [11,12].

Management of glomus tumors entails complete surgical removal of the lesion, proving curative in most cases [12]. Recurrence can occur, often within a year of excision, and is usually attributed to insufficient removal rather than true recurrence [13].

Conclusion

In summary, glomus tumors represent benign soft tissue tumors, with clinical diagnosis typically straightforward in cases of typical localization. However, in atypical locations, imaging, particularly MRI, assumes a pivotal role in establishing the diagnosis. Histological examination serves to confirm the diagnosis. The gradual onset of chest pain after a period of coughing, unresponsive to common analgesics, should prompt consideration of a glomus tumor in an atypical location. The combined use of MRI and surgical intervention, involving complete excision and subsequent histological analysis, facilitates a confirmed diagnosis and ensures remission.

Conflict of Interest: Authors declare no conflict of interest.

References

1. Gombos Z, Zhang PJ (2008) Glomus tumor. Arch Pathol Lab Med 132(9):1448–52.
2. Masson P (1924) Le glomusneuromyo-drterialeds resions tactiles et ses tumeurs. Lyon Chil 21:257-80.
3. Brems H, Park C, Maertens O, Pemov A, Messiaen L, et al. (2009) Glomus tumors in neurofibromatosis type 1: genetic, functional, and clinical evidence of a novel association. Cancer Res 69(18):7393-401.
4. Jalan D, Elhence A, Rathore DS, Maley DK (2016) A recurred subungual glomus tumour of the thumb. BMJ Case Rep 2016:bcr2015212963.
5. El Jouari O, Gallouj S, Elloudi S, Senhaji G, Rimani M, et al. (2018) A painless glomus tumor: a case report. J Med Case Rep 12(1):302.
6. Lattes R, Bull DC (1948) A Case of Glomus Tumor with Primary Involvement of Bone. Ann Surg 127(1):187-91.
7. Mravic M, LaChaud G, Nguyen A, Scott MA, Dry SM, et al. (2015) Clinical and Histopathological Diagnosis of Glomus Tumor: An Institutional Experience of 138 Cases. Int J Surg Pathol 23(3):181-8.
8. Dong-Yeong L, Sun-Chul H, Soon-Taek J, Dae-Cheol N, Jin-Sung P, et al. (2015) The value of diagnostic ultrasonography in the assessment of a glomus tumor of the subcutaneous layer of the forearm mimicking a hemangioma: a case report - PubMed [Internet] J Med Case Rep 9:191
9. Stacy GS, Bonham J, Chang A, Thomas S (2020) Soft-Tissue Tumors of the Hand-Imaging Features. Can Assoc Radiol J 71(2):161-73.
10. Perţea M, Poroch V, Velenciuc N, Mitrea M, Luncă S, et al. (2021) Clinical, histopathological and immunohistochemical features of glomus tumor of the nail bed. Rom J Morphol Embryol 62(1):233-8.
11. Singh V, Kumar V, Singh H, Kakkar N (2020) Primary pulmonary glomus tumour: a diagnostic challenge. BMJ Case Rep 13(2):e231926.
12. Zanjani LO, Shafiee Nia B, Vosoughi F, Mirzaian E, Aghaghazvini L, et al. (2021) An unusual case of chest wall glomus tumor presenting with axillary pain: a case report and literature review. Eur J Med Res 26(1):49.
13. Patel J, Vaish A, Vaishya R, Singhal S (2022) Reappearance of a glomus tumor of the finger after nine years- A rare case report and literature review. Journal of Orthopaedic Reports 1(1):38-41.