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A rare late recurrent glomus tumor of the knee: A case report

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Abstract

Background: Glomus Tumors (GTs) usually occur in young women, most frequently under the nail. Combined with the relevant medical history and unique signs, a preliminary diagnosis can be made, with confirmation by pathological examination. A few cases are seen outside the nail, generally located in the fingers, toes, and beyond the hands and feet, but these are rare. Extra-nail GT is often misdiagnosed and mistreated because of atypical symptoms or "pain point migration." Simultaneously, GT often relapses within a short period due to partial resection, which causes great pain and is a serious economic burden to patients.

Case presentation: Here in, we report a painful subcutaneous mass on the knee of an elderly man, 63 years old, Han nationality. It was resected 10 years prior and recurred more than 8 years after the operation. The pain was very difficult for the patient to endure. Thus, surgical resection was performed, and the scope of resection was expanded during the operation. There was no recurrence at the 3-year follow-up. Accordingly, we propose a new view on the scope of surgical resection.

Conclusion: Generally, GTs should be considered first for local pain in the non-subnail region with constant location, particularly severe pain with tenderness on compression. Complete resection is the key to reducing recurrence. Since the recurrence period can be several years, thorough long-term follow-up should be implemented post-resection.

Keywords: Glomus tumor; Perivascular cell tumor; Late recurrence; Case report.

Abbreviations: GTs: Glomus tumors; HE: Hematoxylin-eosin staining; MRI: Magnetic resonance imaging.

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Background

Glomus Tumors (GTs) are rare benign tumors, accounting for less than 2% of soft tissue tumors, most frequently occurring under the nail [1]. These tumors are easily diagnosed by the typical triad of paroxysmal pain, tenderness, and cold hypersensitivity [2]. However, GTs occurring outside the nail—such as the knee joint—are even rarer, and the symptoms are atypical, leading to misdiagnosis and mismanagement. The tumor is small, and often incomplete resection leads to recurrence [2]. We report a case of an older man who relapsed 8 years after surgery and provide a new perspective on the scope of surgical resection.

Case presentation

A 63-year-old man, presented with recurrence after the resection of a subcutaneous painful mass on the right knee more than 10 years ago. At that time, the patient had a subcutaneous mass on his right knee, about the size of a soybean grain, accompanied by obvious tenderness. Tumor resection was performed in a local hospital without pathological examination (details unknown). The wound healed as expected, there was no recurrence of pain and tumor, and recovery was good. Then, two years ago, on the inner side of the patella of the right knee—where the operation was performed— a blue tumor the size of a soybean grain was identified. The tumor was obviously tender, and even clothing friction could cause severe pain. In the last six months, the tumor had increased in size and the tenderness was even more obvious; therefore, the patient presented at our hospital. The patient had a 10-year history of hypertension and denied foreign body puncture, trauma, diabetes, and gout history. On physical examination, no superficial lymph nodes were palpable, but a dark purple mass the size of a peanut on the medial side of the right patella (Figure 1a) was noted. The skin was unbroken, with no obvious scars, the boundary was still clear, mobility was good, and there was no adhesion to the deep subcutaneous tissue; however, there was obvious tenderness, and the patient resolutely refused touch and pressure by the doctor. The Pin test was positive (i.e., the love test, where there are obvious tenderness points when pressing the tail with a pin). The patient refused repeated examination due to severe pain. Local color Doppler ultrasonography could not be performed, and the tumor was too small for MRI. After considering the superficial and clear boundary of the tumor and communicating with the patient and his family, the patient agreed to undergo surgical resection with postoperative pathological examination for a clear diagnosis.

The outpatient department administered 1% lidocaine containing 1:100,000 adrenaline to infiltrate the area around the tumor. After the anesthesia took effect—about 2 mm away from the edge of the tumor—a shuttle incision was created (Figure 1b), the skin was cut to the deep fascia, separated, and it was noted that the tumor was located on the superficial surface of the deep fascia, with no relationship to small blood vessels and nerves. The tumor, and the surrounding subcutaneous tissue of approximately 2 mm, were completely removed without definite active bleeding and intermittent wound closure was used. The section of the cut specimen showed a "gyrus-like" structure (indicated by the arrow in Figure 1c) with a complete capsule. Postoperative pathological examination showed many round

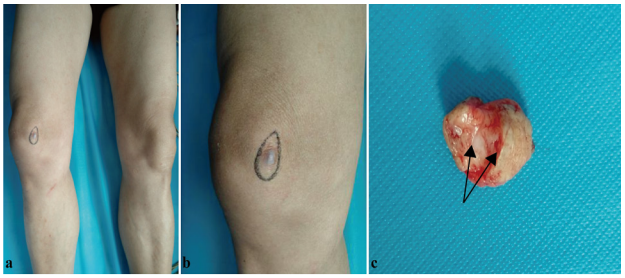

Figure 1: (a) Site of the painful mass.(b) Preoperative markings indicating the resection area. (c) Mass section, arrow indicates the "gyrus-like" structure of the mass section.

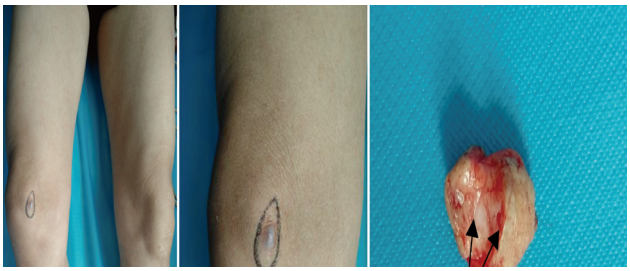

Figure 2: Tumor composed of round cells with uniform size and abundant interstitial blood vessels [Hematoxylin-Eosin staining (HE) × 60, enlarge ×100].

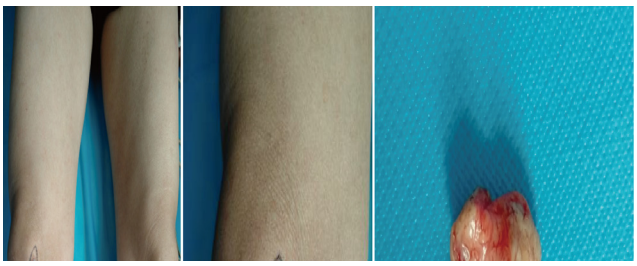

Figure 3: Immunohistochemistry results consistent with bulbous hemangioma. (A): SMA (+ve. (B): CD31 (+ve. (C): CD34 (+ve. (D): CK (-ve.

cells with consistent proliferation, abundant interstitial vessels, no obvious cellular atypia, no definite malignant lesions, and the local tumor base was close to the cutting edge (Figure 2). Immunohistochemistry showed CD31 (vascular+), Desmin (focal+), SMA (+), Ki67 (1% positive), CD34 (vascular+), CK (AE1/AE3) (-), S-100 (-), ERG (vascular+), and CD68 (-), consistent with bulbous vascular-venous malformation (bulbous hemangioma) (Figure 3). One-stage postoperative wound healing occurred. At the 3-year follow-up local pain was eliminated, and there was no tumor recurrence.

Discussion

The glomus bodies—a nerve-smooth muscle device between small arteries and veins—control local blood flow and blood pressure and have a thermoregulatory effect by regulating peripheral vasoconstriction. The vascular bulb is distributed more at the ends of the extremities, particularly in the fingers, palms, toes, and soles of the feet [1]. When this tissue overgrows, a Glomus tumors (GTs) forms. According to the World Health Organization classification, GTs is a well-differentiated, mesenchymal, perivascular, cellular tumor [3]. GTs is usually benign and rarely malignant. GTs is common in women aged 20–50 years,

although extra-nail GTs is more common in men aged 40–70 years [2]. GTs may be related to the underdevelopment of the angiosphere in minors and the degeneration of the angiosphere in the elderly. It occurs mainly in the extremities, with 75% of GTs cases occurring in the hands, with the most common site being the subnail. Other sites, such as the oral and nasal cavities and internal organs, are rarely reported [2]. The tumor is small, usually no more than 1 cm in diameter, often solitary, but occasionally multiple [1]. Because the vascular sphere is often rich in nerve endings, when the tumor encounters stimuli like cold, heat, collision, pressure, even emotional fluctuations, and other factors that affect vasomotor function, there is severe pain. Therefore, GTs has a typical paroxysmal "triad" of severe pain, tenderness, and cold hypersensitivity [1]. The diagnosis can be made using the Love or cold sensitivity test. However, in some parts of the body, such as the leg, two-point resolution is often more than 30 mm, so the pain point indicated by the patient and the pain point examined by the doctor can be tens of millimeters apart. Additionally, the patient presents with "pain point wandering," that is, the pain point cannot be clearly defined, and the patient is afraid of touching the tumor and repeated examination, which can easily mislead the clinician. Particularly, the proportion of missed diagnosis, misdiagnosis, and mistreatment of extra-nail GTs is high. In this older man, the lesion was located on the knee—a rare site. Although the location was atypical, the symptoms were relatively typical, and GTs outside the nail was suspected according to the medical history and clinical manifestations.

Subnail GTs can easily be preliminarily diagnosed based on typical symptoms and signs. However, for extra-nail GTs, the diagnosis is difficult, particularly because the tumor may be small and hidden, the symptoms may be atypical, and there may be a lack of specificity. Thus, an auxiliary examination can be done, when necessary. In a published case report, color ultrasonography showed that the tumor was mostly hypoechoic, and color Doppler showed abundant blood flow signals in or around the tumor, showing a flower ring/small fireball shape, with specificity [4]. Therefore, ultrasound can be used for early diagnosis and treatment of extra-nail GTs. MRI has good resolution with soft tissue, and GTs shows a moderate or low signal on T1WI and a significantly high signal on T2WI [4]. However, for lesions smaller than 3 mm, the sensitivity is lost, and MRI is expensive, so there are difficulties with incorporating it as a routine examination. The final diagnosis requires both pathology and immunohistochemistry. The pathological examination will show a round or oval, dark red or dark purple, clear boundary, complete capsule, middle, blood outflow from the tumor after incision, dark gray tumor. Microscopic examination will show tumor tissue characterized by vascular spheres simulating arteriovenous anastomotic plexuses. There are many thick-walled blood vessels or capillaries in the tumor, and multilayer spheroidal tumor cells are surrounded by blood vessels. The size and morphology of the spheroidal tumor cells are relatively consistent; the nucleus is round or oval, centered, with inconspicuous nucleoli, and sometimes there are unmyelinated nerve fibers traveling through the space between the tumor tissues. There are different numbers of mast cells and lymphocyte infiltrates, and the surrounding intact capsule is seen [5]. According to the relative proportion of spherical cells, blood vessels, and smooth muscle, GTs can be divided into Solid bulbous tumor, the most common subtype, accounting for about 75%. Glomus hemangioma, which makes up about 20%, and Glomus leiomyoma, accounting for less than 10% [1]. Immunohistochemistry will show vimentin, SMA (+), indicating that the tumor cells are myogenic, CD31 (+) indicating that the tumor cells are vasogenic, and partial expression of CD34; however, desmin, CK, Syn, Ki67, and S-100 are generally negative, [6] which is helpful in the differential diagnosis. The tumor in our patient was a bulbar hemangioma. This was determined based on the pathological characteristics, such as SMA (+), CD31 (vascular+), Desmin (focal+), CD34 (vascular+), ERG (vascular+), CK (AE1/AE3) (-), S-100 (-), CD68 (-).

GTs is rare in the clinic and is mainly differentiated from other tumors by pain. Malignant glomus tumors are extremely rare, and the main features are as follows: The tumor is deep, more than 2 cm in diameter, and located under fascia or viscera; or an atypical mitotic image can be seen; or the nucleus has obvious atypia and different degrees of mitotic activity [7]. Blue rubber bled nevus syndrome has multiple skin lesions and a lack of glomus cells in histopathology. Neuroma is seen at the site of trauma, operation, or amputation, and clinically it is mostly an isolated solid subcutaneous nodule with normal or light brown skin color. A neuroma is easy to distinguish from the abovementioned diseases.

Surgical resection is the only effective treatment for GTs, with either preoperative diagnosis or high suspicion. However, due to the small size of the tumor, it is often difficult to remove the tumor or find the tumor tissue, which can lead to postoperative recurrence. Thus, the recurrence rate is about 12%–33%. Early recurrence (days to weeks after the operation) is usually due to incomplete resection or undiagnosed secondary tumors. Delayed recurrence (2–3 years after the operation) is mainly due to newly developed undetected tumors or synchronous satellite lesions [2,8]. In our patient, the tumor was located subcutaneously at the knee joint, which is a rare site and prone to misdiagnosis; he had previously undergone tumor resection. On careful inquiry about the patient's medical history, we postulated that although the recurrence interval was long in this patient, it was caused by incomplete resection, and the location was the same. More than 8 years after the first resection, we removed the tumor and its surrounding subcutaneous tissue completely. The tumor was diagnosed as a glomus tumor by postoperative pathological examination. After 3 years of follow-up, there was no tumor recurrence. We postulated that the recurrence in this patient was caused by incomplete resection, although the recurrence interval was long. This was possibly because the location of the lesion was hidden, the early symptoms were not obvious, and it was not found in the early stage. Thus, clinicians should be vigilant and pay attention during long-term follow-ups post resection.

Conclusion

Extra-nail GTs is a rare benign tumor originating from mesenchymal tissue, which often seriously affects the quality of life of patients due to pain. For this type of GTs, clinicians should understand its importance; make a timely, correct diagnosis; and implement extended resection by removing as much of the diseased tissue as possible or removing it with more than 1 mm of the surrounding normal tissue. If necessary, microsurgical technology should be used to completely remove the tumor, to avoid recurrence and reduce patient pain.

Declarations

Ethics approval and consent to participate: This study was approved by the Research Ethics Committee of the Second Hos-

pital of Jilin University.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Availability of data and materials: All data generated or analyzed during this study are included in article.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: XLO wrote the manuscript and reviewed the clinical notes; RL and WRQ revised the manuscript and confirmed the histopathological examination results; HT analyzed and interpreted the patient data and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

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References

1. Gombos Z, Zhang PJ. Glomus tumor. Arch Pathol Lab Med. 2008; 132: 1448-1452.
2. Schiefer TK, Parker WL, Amadio PC, Inwards CY, Spinner RJ, et al. Extradigital glomus tumors: A 20-year experience. Mayo Clinic Proceedings. 2006; 81: 1337-1344.
3. Kallen ME, Hornick JL. The 2020 WHO Classification: What's New in Soft Tissue Tumor Pathology? American Journal of Surgical Pathology. 2020; Publish Ahead of Print.
4. Baek HJ, Sun JL, Cho KH, Choo HJ, Lee SM, et al. Subungual tumors: Clinicopathologic correlation with US and MR imaging findings. Radiographics. 2010; 30: 1621-1636.
5. Tsuneyoshi M, Enjoji M. Glomus tumor: A clinicopathologic and electron microscopic study. Cancer. 1982; 50: 1601-1607.
6. Folpe AL, Fanburg–Smith JC, Miettinen M, Weiss SW. Atypical and Malignant Glomus Tumors: Analysis of 52 Cases, With a Proposal for the Reclassification of Glomus Tumors. American Journal of Surgical Pathology. 2001; 25: 1-12.
7. Fletcher C. The evolving classification of soft tissue tumours: An update based on the new WHO classification. Histopathology. 2013; 64: 2-11.
8. Luzar B, Martin B, Fisher C, Calonje E. Cutaneous malignant glomus tumours: Applicability of currently established malignancy criteria for tumours occurring in the skin. Pathology. 2018; 50.