CASE REPORT

A recurred subungual glomus tumour of the thumb

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SUMMARY

Glomus tumours are rare benign neoplasms arising from the glomus body, accounting for 1–5% of all soft-tissue tumours of the hand. The diagnosis of glomus tumour is frequently delayed, varying from 3 to 15 years. We present a case of a recurred glomus tumour of the thumb where symptoms persisted for a period of 6 years after an attempted surgical excision. The patient presented with classical symptoms of localised tenderness, cold sensitivity and excruciating pain on touch. Findings on radiograph, ultrasonography and contrast-enhanced MRI further supported the diagnosis. Complete surgical excision, the only recommended treatment, resulted in complete symptomatic relief to the patient. This case report highlights the importance of meticulous complete excision and clinical awareness of the disease among primary care physicians.

BACKGROUND

Glomus tumours are rare benign neoplasms arising from the glomus body, which controls blood pressure and temperature by regulating peripheral cutaneous blood flow.^{1–3} These tumours account for 1–5% of all soft-tissue tumours of the hand.⁴ Fifty to Seventy-five per cent of all glomus tumours have shown a predilection to occur in the hand.¹ The diagnosis of glomus tumour is frequently delayed because of the low prevalence and lack of awareness of the condition among primary care physicians.^{5–6} Delay in diagnosis is between 3 and 15 years in various studies.^{7–8} Recurrence postexcision is probably more common than originally documented and ranges from 5% to 15%.^{1–6–9–10} We present a case of a recurred glomus tumour of the thumb where symptoms persisted for a period of 6 years after an attempted surgical excision.

CASE PRESENTATION

A 45-year-old woman presented with a 7-year history of excruciating pain in the left thumb provoked by even the slightest touch. She reported of intolerance to cold in the same digit. She had undergone surgery at another centre for this problem 6 years prior, with no relief in symptoms. She had consulted several primary care physicians and was advised analgesics, off and on, without permanent relief of symptoms. There was no history of trauma, no fever and no involvement of the other joints.

Examination revealed a bluish discolouration and an accompanying nodular swelling at the base of the nail plate of the left thumb. The nail plate showed mild deformation probably as a result of the pressure effect of the underlying tumour or scarring due to the previous surgery (figure 1). The

swelling was tender to the touch. Love's pin test was found to be positive. Movements at the distal interphalangeal (DIP) joint were preserved.

INVESTIGATIONS

Plain radiograph of the left thumb showed scalloping of the dorsal surface of the distal phalanx (figure 2). Ultrasonography revealed a well-defined oval shaped predominantly hypoechoic soft tissue density mass lesion measuring about 7×7 mm over the dorsal aspect of the distal phalanx of the left thumb, with no evidence of vascularity within the lesion (figure 3). Contrast-enhanced MRI of the left thumb revealed a well-defined intensely enhancing solid lesion on the extensor surface of the left thumb abutting the distal phalanx. The lesion appeared hyperintense on T2-weighted (T2W) and isointense on T1-weighted (T1W) images. There was cortical scalloping of the extensor surface of the shaft of the distal phalanx, with neither osseous destruction nor marrow oedema (figure 4A–C).

DIFFERENTIAL DIAGNOSIS

The differential diagnoses considered were glomus tumour, subungual neuroma and subungual exostosis.

The presentation of the patient, including age and female sex, along with the characteristic triad of exquisite pain, localised tenderness and cold intolerance, favoured the diagnosis of glomus tumour. The imaging studies including ultrasound and MRI further supported the diagnosis.

The second differential considered, subungual neuroma, can also cause sharp, excruciating pain in the finger when touched, but it is usually post-traumatic or iatrogenic.^{11–13} On ultrasonography, hypoechoic and poorly defined solid tumours can be seen, but MRI helps to characterise these

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Figure 1 Preoperative clinical photograph of the left thumb.

Figure 2 Radiograph of the left thumb showing scalloping on the dorsal aspect of the distal phalanx.

tumours and to identify the neurovascular bundle, and thereby the nerves with which they are associated.¹⁴ The MRI findings were against the diagnosis of neuroma.

Finally, subungual exostoses are tumours that usually arise underneath the nail bed and have a similar presentation. However, the imaging studies were against the diagnosis of subungual exostosis.¹⁵

TREATMENT

The above differential led to a decision to perform an excisional biopsy.

After obtaining informed consent, an excision biopsy was performed using a direct transungual approach. The choice of approach was decided on the basis that, as this was a recurrent lesion, more extensive exposure was required, which would not be possible using a lateral approach or other minimally invasive approaches. Second, the major disadvantage of using a transungual approach is nail deformity, which was already present in our case because of previous surgery.² Intraoperatively, after

removing the nail plate, a bluish red 7 mm diameter tumour (figure 5A–C) was exposed and excised completely. The excised tumour (figure 6) was then sent for histopathological confirmation.

OUTCOME AND FOLLOW-UP

Histopathological examination (figure 7) confirmed the diagnosis of glomus tumour. The postoperative period was uneventful and the patient's symptoms were relieved immediately after the surgery. She was followed up at 2 weeks, 6 weeks and thereafter at 3-month intervals for 1.5 years. At the latest follow-up at 1.5 years, the patient was asymptomatic with no evidence of recurrence (figure 8), and very satisfied with the cosmetic result as well as with the clinical resolution of her symptoms. The nail plate changes after the surgical procedure gradually reduced to just a linear ridge over the nail plate extending from the proximal nail fold to the distal tip.

DISCUSSION

Glomus tumour was first described by Wood, in 1812, and the histological description was given in 1924 by Barre and Masson.^{16 17} The tumour is usually more common in women and occurs most frequently in the subungual region of the distal phalanx.^{9 18} These tumours arise from the normal glomus apparatus, located in the subcutaneous tissue, especially in the tips of the fingers. Glomus tumours are classified as either solitary or multiple. The majority of cases are solitary tumours, most frequently seen in the fingertips. They are encapsulated, usually subungual, and contain numerous small vascular lumina. In contrast, multiple tumours are non-encapsulated, rarely subungual and have larger vascular spaces. In addition, multiple glomus tumours are often asymptomatic and present earlier in life.¹⁹

Clinically, glomus tumours are characterised by a triad of localised tenderness, cold sensitivity and excruciating pain on touch. Despite a classical history and examination, the reported delay in diagnosis is up to 15 years due to lack of awareness of the condition among primary care physicians.^{7 8 20} Fazwi *et al*,⁶ in their study, stated that a glomus tumour was diagnosed much earlier by the resident hand surgeon and with increasing clinical experience, thereby highlighting the importance of awareness of the disease among the primary care physicians.

Various clinical tests for glomus tumour include a Love's pin test (100% sensitivity, 0% specificity and 78% accuracy),

Figure 3 Ultrasound image showing a well-defined oval shaped predominantly hypoechoic soft tissue density mass lesion measuring about 7×7 mm.

Figure 4 MRI of the distal phalanx showing a well-defined solid lesion on the extensor surface of the left thumb abutting the distal phalanx appearing isointense on T1-weighted image (A), hyperintense on T2-weighted image (B) and postcontrast enhancement (C).

Hildreth's test (71.4–92% sensitivity, 91–100% specificity and 78% accuracy) and a cold sensitivity test (100% sensitivity, specificity and accuracy).^{2 21 22}

Plain radiographs are normal in the majority of cases. Long-standing cases with large tumours (22% of cases), however, may show scalloping of the distal phalanx, as was evident in the present case.²³

Ultrasonography helps in confirming the site and determining the shape and size of the tumour (as small as 3 mm), preoperatively.^{24 25} Colour duplex Doppler sonography is preferred and helps in differentiating the tumour from other hypoechoic subungual tumours.^{25 26}

MRI is the most sensitive imaging modality available, with a sensitivity of 90%, specificity of 50%, and positive predictive value of 97% and negative predictive value of 20%.^{18 27} The tumour classically has low signal intensity on T1W images and high signal intensity on T2W images, with strong enhancement on postcontrast images.^{19 27} However, its role in diagnosing glomus tumour remains controversial. As stated in the recent literature, non-diagnostic MRI is associated with atypical pathology, atypical location, absence of bone erosion and lack of clinical suspicion, thereby highlighting the importance of clinical suspicion in diagnosing the tumour.²⁸ The literature documents the fact that an excisional exploration may still be performed on a strong clinical suspicion, even if the non-contrast MRI scan does not support the diagnosis of a glomus tumour.²⁷ Nonetheless, MRI aids in preoperative surgical planning by

Figure 5 Intraoperative images of the glomus tumour. (A) Bluish discolouration after removal of the nail plate. (B) Rounded gelatinous mass after incising the nail bed. (C) Showing complete excision and scalloping over the dorsal surface of the distal phalanx.

Figure 6 Gross appearance of the excised glomus tumour measuring 0.7×0.7 cm.

helping to assess the precise location, size and multifocal occurrences and hence preventing recurrences.^{18 27 28}

Complete surgical excision is curative.^{1 2} The literature describes several surgical approaches, each with its own set of advantages and limitations. The traditional transungual approach affords excellent exposure of the centrally placed tumours, however, chances of postoperative nail deformity are high.^{2 29–31} In our case, the above approach was used, as it was a recurrent tumour following an attempted surgical excision, and some amount of nail deformity was already present in the case. The Keyser-Littler approach is through a high mid-lateral incision below the lateral skin fold, however, it compromises the interosseous structures that form the lateral support for the nail matrix and nail plate.^{2 10} In a lateral subperiosteal approach, the tumour is accessed without retracting the interosseous structures, however, the degree of exposure is limited.¹⁰ A modified periungual approach involving an L-shaped incision was described by Fong *et al.*³¹ Although this approach did not require splitting of the nail bed, it was found to be suitable only for peripheral tumours. Wang *et al* described a nail bed margin approach, where a curved incision was made near the side of the tumour along the nail bed margin. According to the authors, it provided good exposure of the tumour at all subungual locations while avoiding postoperative nail deformity.³² Another innovative approach was described in which the nail plate and nail bed were transected with a single incision to expose the tumour, so as to reduce trauma to the nail bed.²⁹

Various authors have reported a recurrence rate of 4–15%;^{1 6 9} the recent literature, however, states that recurrence rates may actually be as high as 50%.³³

True recurrence is observed within 1 year of surgery and is usually indicative of inadequate excision, as was documented in the present case.³⁴ Carroll and Berman stated, in their series of

Figure 7 Encapsulated tumour composed of blood vessels surrounded by nests and islands of round cells with eosinophilic cytoplasm in a myxoid stroma (H&E stain, ×40).

28 cases of glomus tumours, that “meticulous complete excision, the only recommended treatment, should result in complete cure”, and that “recurrent symptoms should suggest incomplete excision rather than a recurrence.”¹

Delayed recurrences presenting after 1 year, are believed to be due to concurrent satellite lesions that are missed at the time of index surgery, resulting in the development of a new tumour near the site of excision.^{3 17} In fact, case reports of multiple synchronous glomus tumours discovered intraoperatively seem to support this assertion.³⁵

Figure 8 Clinical photograph of the left thumb at 1.5 years follow-up.

Learning points

- ▶ Glomus tumours, despite a classical history and examination, have a reported delay in diagnosis for up to 15 years, which can be improved by increasing the awareness of the disease among primary care physicians.
- ▶ MRI is the most sensitive imaging modality available, with a sensitivity of 90% and positive predictive value of 97%.
- ▶ Meticulous complete excision, the only recommended treatment, results in complete cure; and early recurrence/persistence of symptoms indicates incomplete excision.

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