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Glomus Tumor of the Fingers: Accurate Preoperative Localization as a Prerequisite to Avoid Tumor Recurrence

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ABSTRACT

Glomus tumor (glomangioma) is a rare benign slowly growing exquisitely painful tumor constituting about 2% of all hand tumors. It arises from a neuromyoarterial glomus, which is an arteriovenous anastomosis functioning without an intermediary capillary bed. Normal glomus bodies are found in the dermal retinacular layer of the skin and thought to aid in the thermoregulation of skin circulation and to be highly concentrated in the finger tips, particularly beneath the nail bed **[1]**. Glomus tumors that was described first by Masson in 1942, can occur anywhere in the skin or soft tissue, and the most common site is the finger **[3]**. The tumor usually presents as a painful, firm, purplish solitary nodule of the extremities, most commonly in the nail bed **[2]**.

Preoperative tumor localization plays crucial role in the surgical treatment of glomus tumor without recurrence. Depending on the clinical triade, provocative clinical tests and MR imaging proper tumor site can be detected with high precision. The study comprised 20 patients with tender finger tip, nail ridging and blue spots for variable durations. The transungular access route was preferred as 90% of the cases were presented with centrally located tumors. The postoperative nail dystrophy encountered in 2 patients treated with transungular route and recurrence took place in two patients. In conclusion, glomus tumor is rare but disabled tumor that should be suspected in patients suffering from chronic finger tip pain. The main diagnostic tool after clinical examination is MRI which help in detecting the actual tumor location for complete tumor excision without recurrence.

INTRODUCTION

Glomus tumor (glomangioma) is a rare benign slowly growing exquisitely painful tumor constituting about 2% of all hand tumors. It arises from a neuromyoarterial glomus, which is an arteriovenous anastomosis functioning without an intermediary capillary bed. Normal glomus bodies are found in the dermal retinacular layer of the skin and thought to aid in the thermoregulation of skin circulation and to be highly concentrated in the finger tips, particularly beneath the nail bed **[1]**. Glomus tumors that was described first by Masson in 1942, can occur anywhere in the skin or soft tissue, and the most common site is the finger **[3]**. The tumor usually presents as a painful, firm, purplish solitary nodule of the extremities, most commonly in the nail bed **[2]**.

However, it is difficult to diagnose because of obscure symptoms, such as chronic pain and hypersensitivity, and the small size of the lesion. This tumor has solitary and multiple forms. Solitary tumors are commonly found in the distal phalanx in a paraungual or subungual location. They present with a classic triad of paroxysmal pain, pinpoint tenderness, and cold hypersensitivity. Multiple tumors are extremely rare, are inherited in an autosomal-dominant pattern, and can have involvement outside the hand and may be associated with von Recklinghausen' s neurofibromatosis. Histopathologic characteristics can be used to differentiate the solitary and multiple forms **[4-6]**.

Glomus tumors are usually located in the distal segment of the fingers under the nail matrix or in the pulp. Symptoms may have been persisting for a long time because of failure in diagnosis and treatment. Changes in temperature, palpation, and touching may cause pain and hypersensitivity **[7]**. When located in deep layers like in pulp, it is difficult to make a diagnosis because of less obvious hypersensitivity, there is no palpable mass and the extent of hypersensitive area is almost a pencil point wide. These patients without objective findings usually do not accept an operation **[8]**. Transillumination **[9]**, MRI **[10]** and US **[7]** are useful tools to confirm the tumor location. As long as there is no bony erosion, plain X-rays can not be helpful except in long standing cases where bony erosions are evident **[11]**.

The aim of the present study is to shed light on rare but debilitating tumor and emphasize the role of proper preoperative tumor site localization that

leads to successful surgical excision and avoid tumor recurrence.

PATIENTS AND METHODS

The study comprised 20 patients presented to the Plastic Surgery Unit of Alexandria University during the period from October 2005-February 2010. Fifteen patients were females and 5 were males, their ages ranged from 24-60 years with an average of 35 years. The presenting complaint was spontaneous pain, nail ridging, splitting and localized point of tenderness at the finger tip. Time between onset of symptoms and diagnosis ranged from 4 months to 3 years. Diagnosis was suspected preoperatively in all cases based on clinical and radiological examinations. Diagnostic clinical tests included Love' sign, Hildreth' sign and transillumination. Love' sign i.e. pressure applied over the lesion with the tip of a pencil eliciting excruciating pain [12]. Hildreth' sign i.e. the point pain diminished after slowly insufflating a brachial cuff [13].

All patients were subjected to:

1- Full history taking, clinical examination (local and general) routine laboratory investigations.

2- Medical Imaging studies including:

A- plain X-ray for hand A-P (anteroposterior) and lateral views.

B- MR imaging of the involved finger was performed as a confirmatory tool and to detect actual tumor location.

MRI was performed using 1.0 tesla closed magnet (Philips Machine Unit) at the Radiodiagnosis Department of Alexandria Main University Hospital. The following sequences were obtained in coronal, sagittal and axial planes:

- T1-weighted image spin-echo sequence.
- T2-weighted image spin-echo sequence.
- Post contrast T1-weighted spin echo after I-V injection of Gadolinium contrast medium 2mmol/kg body weight.

Plain X-ray revealed positive changes only in long standing cases in the form of bone notching or indentation of the distal phalanx and bone thickening (Fig. 1). Magnetic resonance (MRI) typical image was a low signal on T1 and a high signal on T2. A small space occupying lesion was detected at the subungual area and adjacent to the distal phalangeal bone. The mass was isointense of the dermis of the nail bed on T1-weighted image and hyperintense on T2-weighted image (Fig. 2A,B). The post contrast-enhanced image of the middle finger showed homogenous contrast uptake and high signal intensity on T1-weighted sequence (Fig. 3A,B).

Surgical intervention:

The operations were performed under local anesthesia, in a bloodless field using arm tourniquet and with the aid of magnifying loupes. MR images must be examined carefully preoperatively to decide the proper approach depending on the exact tumor location. In central (subungual) lesions, a direct transungular approach was performed by removing the nail plate and incising the nail bed longitudinally to explore the underneath tumor. In peripheral lesions, a lateroungual approach was preferred, where a lateral incision was done close to and parallel to the nail margin accompanied with partial nail removal if necessary. Always explore the whole area for any tiny tumors that may be missed out. After ensuring complete extirpation of the tumor within its capsule, meticulous direct closure of the nail bed was done using fine 7/0 or 8/0 absorpable suture material. Then, the wound was covered with non adhesive gauze dressing for 1-2 weeks. All excised tumor tissues were examined histopathologically. Patients were followed-up for a period of 3 months -4 years to look for possible recurrences and residual nail deformities.

RESULTS

Follow-up in this study ranged from 4 months-4 years. Clinical diagnosis was accurate in all patients using the classic triad of pain, tenderness and thermal changes and confirmed by the clincal tests described before, MRI examinations and histopathological studies. All patients had complete relief of pain immediately postoperatively. All patients presented by a severely painful finger tip while nail ridging noted in 15 patients and blue spots in 5 patients. Right hand lesions were encountered in 14 patients and left hand in 6 patients, 5 cases with thumb lesions, 6 cases with index, 6 cases with ring finger and 3 with middle finger lesions. Eighteen patients with sububgual lesions 90% (nail bed area) and 2 patients with peripheral lesions 10% (nail fold).

Preoperative tumor site localization was accurately located using the clinical tests and radiological studies. Hildreth' test was sensitive in 16 patients (80%) and Love's test in 100% of the patients. Plain X-ray showed bone thickening and indentation in only three patients with long standing presentation (15%). MRI was effective in accurately locating the tumor site in 18 patients (90%) and was doubtful in 2 patients with subungual location

as the patients had old trauma to the nail bed area which caused some picture artifact.

In the patients with doubtful MRI images, tumor location was determined by meticulous clinical diagnosis and provocative clinical tests before surgical exploration was done. Histopathologic studies confirmed the diagnosis of glomus tumor in all patients.

Postoperatively, eighteen patients (90%) were asymptomatic in the follow-up period for 4 years, and in two patients (10%) the symptoms recurred again, after one month in one patient and 2 years later in the other. Re-exploration after second MRI examination revealed a volar extension of the primary tumor which was difficult to explore via transungular route in the patient with early recurrence. In the other patient with late recurrence, the tumor was located away from the original tumor site i.e. a new growth. Postoperative nail ridging was encountered in two patients of the transungual route (10%), whereas no complications encountered in the lateroungual approach patients. Recovery period 2-4 weeks. Tumor size ranged from 2-6mm.

Fig. (1): Plain X-ray (lateral view) of left index finger showing bony indentation of the distal phalanx.

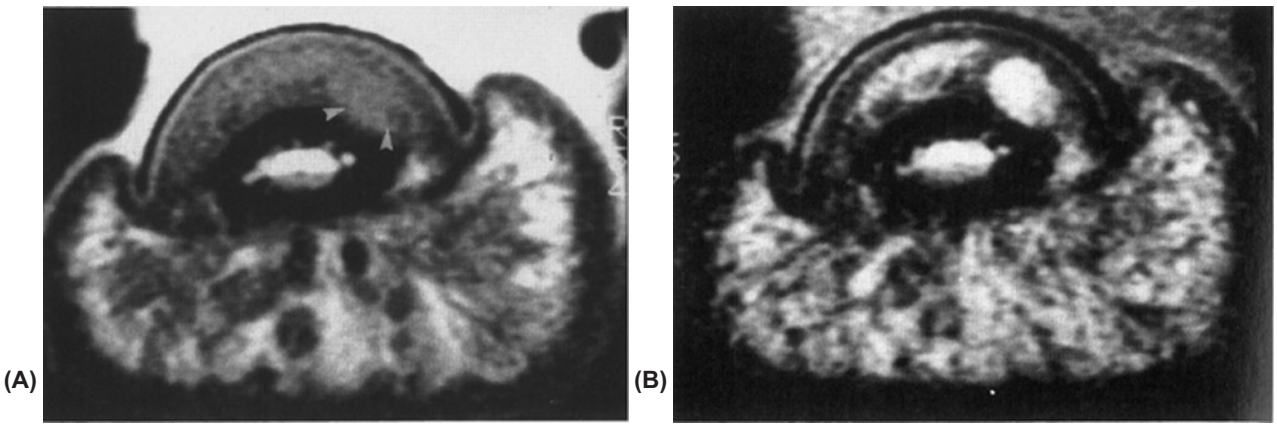

Fig. (2): MR image of glomus tumor at the distal phalanx, a mass is detected at the subungual area (2 small arrows) A) Isointense mass on T1-weighted image and B) Hyperintense mass on T2-weighted image.

Fig. (3): Contrast-enhanced, coronal MR image, T1-weighted sequence (A) and sagittal T2-weighted (B): The arrow pointing to small nodule on the dorsal aspect of the middle finger showing homogenous contrast uptake and high signal intensity on T2-weighted sequence.

Fig. (4-A): Case 1 right thumb with subungual lesion.

Fig. (4-B): Total nail extraction.

Fig. (4-C): Tumor totally excised.

Fig. (4-D): One month postoperatively.

Fig. (5-A): Case 2 left ring finger nail splitting and blue spot.

Fig. (5-B): Total nail extracted and tumor excision.

Fig. (6-A): Case 3 right ring finger subungual lesion.

Fig. (7-A): Case 4 left middle finger with nail ridging.

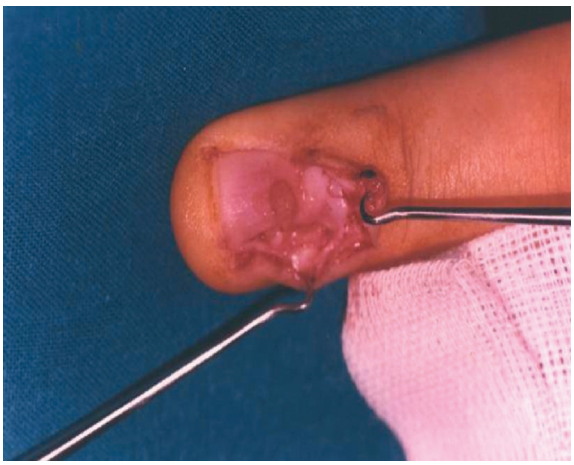

Fig. (6-B): Intraoperative exploration.

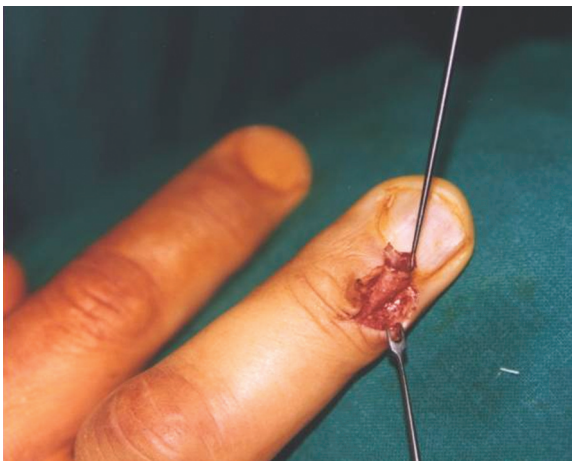

Fig. (7-B): Lateroungual approach with intact nail.

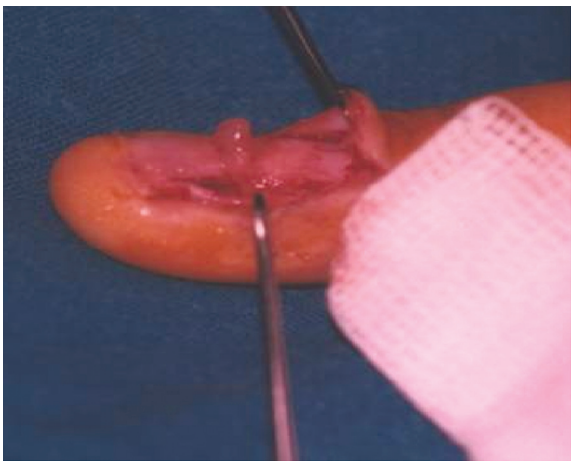

Fig. (6-C): Tumor isolation.

Fig. (7-C): One month post operative.

DISCUSSION

The glomus tumor is a benign tumor usually localized in the distal phalanges of the hand, preferentially in the subungual tissues. Glomus tumor is usually too deep to be easily palpable. Therefore in the presence of clinical triad (pain, tenderness and thermal changes), glomus tumor should be suspected. In addition to the clinical triad, familial inheritance and distal phalangeal localization should direct the clinician towards the diagnosis. However definitive diagnosis can only be established by histologic examination **[4]**.

The reason of the high rate of failed diagnosis and treatment attempts is lack of suspicion during

examination of the patient with impalpable glomus tumor. The fact that it is a rare tumor constituting only about 2% of all hand tumors, may explain the long delay before the right diagnosis is made and therapy is established [4].

Just as it is difficult to convince the patient with impalpable mass to take an operation, so too it is difficult to assess the localization of the tumor during the surgery [4]. If there is discoloration on the nail and palpable sensitive mass, it is easy to determine the location of tumor. It is important to know the tumor location and size for avoiding incomplete excision which is the most important factor in recurrence.

As in many published series the lesion occurred preferentially in middle-aged women, this was in accordance with the presenting study where young female patients constituted 75% of the cases. The preoperative diagnosis can be made from the history, clinical examination and specific tests. Pain triggered by pressure, less often by cold was characteristic. A blue spot under the nail should be searched for in the subungual localization. In the present study, the preoperative clinical tests helped to a great extent in the precise tumor site localization. This was in accordance with study of Raimbeau where Love's test was rarely negative and Hildreth's test was generally contributive for tumor site localization [12,13,14].

Radiological investigations in addition to clinical examinations appear to be essential for the accurate delineation of the extent of the lesions. Plain X-rays can depict glomus tumors, but they have low diagnostic sensitivity for this tumor and do not allow differentiation between recurrent and primary bone erosions [15]. The low percentage of positive radiographs is due to the small size of most lesions and the lesion itself is generally not visible on radiographs [16].

In some series, 25% of the radiographs showed bone erosion, compared with 14-60% in other reports [4,17,18]. In our study, plain X-rays showed bony erosions in only three cases with long standing presentations (15%). These negative radiographs in 17 patients (85%) were attributed to the early presentation of our cases where there was no enough time to develop bony changes. MR imaging however, depicted bone erosions 3.5 times more often than radiography. MR is sensitive for imaging glomus tumor, lesions as small as 1 to 2mm can be detected with high resolution techniques [10]. Glomus tumors are best seen on axial MR image, with low signal intensity on T1-weighted images that is indistinguishable from the nail bed. However, they appear as hyperintense, punctuate foci distinct from the nail bed on both T2 and contrast-enhanced T1-weighted images [19]. It can help differentiate glomus tumors from mucoid cysts and epithelial inclusion cysts because the latter two are non-enhancing. MR image can be particularly helpful when postoperative recurrence is suspected.

MR imaging has become the imaging modality of choice when evaluating soft tissue masses. However, MR imaging descriptions of glomus tumors are rare in the English literature [18,20]. Determination of the exact tumor location is essential to avoid recurrence and nail dystrophy after surgery. Gandon et al. [21] in their series of 48 patients, found the tumors to be located predominantly in the lateral fold, while in our study we noted the subungual location to be the most common site (18 patients i.e. 90%). This is in accordance with the series of Jean-Luc et al. [10] where subungual glomus constituted 85% of their cases.

MR imaging allows the detection and delineation of glomus tumors as well as detection of recurrent glomus [5,22]. In the present study, MRI revealed primary glomus tumor in 18 patients and recurrent tumor in two cases, the smallest tumor size detected was 2mm in diameter.

Other imaging modalities such as ultrasound performed with a high-frequency transducer can detect tumors as small as 3mm in diameter, but the limits of small and flattened subungual lesions are difficult to detect [7,23,24]. In our study, ultrasonography was not used because of its low sensitivity, difficulty in detecting such small lesions and it is operator and techniques dependant. Arteriography [25], thermography, and scintigraphy [26], which has been advocated by some authors, are no longer indicated because of their invasiveness or their poor sensitivity.

The only treatment of primary glomus tumor is surgical removal. The surgical approach depends on an accurate evaluation of the tumor location. Excision is usually curative, although the pain may take several weeks to disappear [27]. The access route used for subungual glomus tumors has been widely debated. Some authors prefer the direct transungular route as it allows easy access without traumatic dissection provided accurate preoperative tumor localization is done [9,28] while others prefer the lateral route as it allowed successful tumor removal whether under the nail bed or those in the nail root as well as those in the lateral ligament of the distal phalanx [15,29,30].

The main surgical approach in the present study was transungual route as the tumors were subungually located in 18 patients. This approach allowed good exploration and easy access to the lesion but the main drawback was the possibility of post operative nail dystrophy. However, with gentle dissection and good alignment of the sutured nail plate, nail dystrophy was reported in only 2 patients (10%).

Moreover, the lateral route allows wide exploration that remains necessary for multiple tumors and even allows an exploration of the pulp to exclude volar extension **[31,32,33]**. With the lateral route the nail plate is not detached avoiding the risk of nail dystrophy which has been reported by other authors preferring the direct access **[15,28,34]**. In the present study, we used the lateral route only in two patients with nail fold lesions and we had no postoperative nail dystrophy. But we encountered this complication only in two patients of our transungual route.

Recurrence of symptoms and the need for repeat surgery have been reported in 12-24% of cases. The time between surgery and MR imaging was 3 and 7 years, respectively **[4]**. MR imaging could be essential in detecting a local extension through an incomplete capsule or multiple locations. Most authors assumed that recurrence is due to inadequate excision, promoting some to recommend more extensive en bloc excision. Regardless of which approach is used, recurrence rates in the digits range from 5%-50% **[4,15,35]**. Higher recurrence rates are related to incomplete excision of the original tumor or to another tumor going unrecognized at the time of initial excision. If recurrence occurs within weeks to months (early) it is the result of inadequate excision, if it is delayed for years, it is likely the result of a new solitary glomus tumor **[36,37]**. Foucher et al., reported 7% recurrence (4 cases) after three to five years **[30]**.

Delayed "recurrence" is possibly due to the development of a new glomus tumor near the excision site **[16,29,38,39]**. In some series, the number of patients with early recurrence was nearly equal to the number of patients with delayed recurrence, with a slight predominance of early recurrences (54%) **[28]**.

In the present study, recurrence occurred in two cases (10%) treated with transungular approach, after one month and 2 years successively. MR imaging studies revealed a glomus tumor near previous surgical scar in the early recurrence case and a new glomus tumor in the late recurrence case. Exploration via lateral approach revealed a volar extension of the primary tumor in the patient with early recurrence and a new growth in the late recurrence. Thereafter, we used MRI study in the recurrent and doubtful cases and was helpful in accurate tumor localization, that lowered the incidence of actual tumor recurrence.

It is suggested that glomus tumors should be kept in mind when considering differential diagnosis of any painful condition of the terminal digit **[40,41]**. However, this benign condition has an unusually high morbidity to the patient before the correct diagnosis is made. This attests to the difficulty in correctly diagnosing this lesion initially. Although history and carefully performed physical examination significantly narrow the differential diagnosis, the patient radiographs are minimally helpful until the bony erosion occurs at the later stages of the disease. With increased index of suspicion, carefully performed history and clinical examination, along with findings on MRI, the treating surgeon can significantly decrease the preoperative morbidity to the patient with glomus tumor **[42,43]**.

Conclusion:

Glomic tumors are certainly uncommon but not exceptional. They should be suspected in patients with digital pain with no apparent cause. The diagnosis is clinical and complementary exploration should be limited to difficult, recurrent or multiple procedure cases. Accurate preoperative tumor site localization is essential in choosing the proper approach and to avoid possible tumor recurrence. Lateral access is ideal for tumors in the nail fold region, allowing excision and exploration for multiple tumors. Direct access can be used in accurately located subungual tumors and with good magnification and gentle dissection complete tumor excision without recurrence can be achieved.

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