Solitary Type of Glomus Tumor Developed in Multiple Sites

– Report of 3 Cases –

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Glomus tumors are usually divided into two types : the classical, tender, painful, solitary type ; and multiple nontender, painless glomangiomas. We report two cases of the solitary type of glomus tumor which developed in multiple nail beds and a case of two solitary type of glomus tumor developed in one nail bed. All three patients presented severe episodic pain and sensitivity to cold, and also showed erythematous to bluish tinge through the nail plates. These tumors revealed the characteristic clinical and histopathologic fingings of the solitary type of glomus tumor except that they developed in multiple sites. So these cases show that the solitary type of glomus tumor can occur in multiple sites. We suggest that they may be some variant of the solitary type of glomus tumor.

(Ann Dermatol 6:(2) 225-229, 1994)

Key Words: Multiple sites Solitary glomus tumor

The glomus tumor, a benign neoplasm derived from a normal glomus, can be solitary or multiple[1]. The more common solitary type usually occurs as a solitary, painful and tender papule on the extremity, particularly in the nail bed, while the multiple type is painless and varies in the number of lesions[2,3,4]. Also histologically, the former can be easily differentiated with the latter in that the solitary type has a fibrous capsule, a smaller vascular space and a larger number of glomus cell layers than the multiple type[5].

Generally the solitary type of glomus tumor is known to develop singly. Only a few cases of the solitary of glomus tumor in multiple sites have been reported in English literature[6,7,8]. But they were not categorized into a specific group.

We report herein two cases of a solitary type of glomus tumor developed in multiple nail beds and a case of two solitary type of glomus tumors in one nail bed, all of which showed typical clinical and histopathologic findings of the solitary type. We suggest that the solitary type of glomus tumor also develops in multiple sites and our cases be taken consideration into the variants of the solitary type.

Received October 4, 1993.
Accepted for publication January 10, 1994.
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REPORT OF CASES

Case 1. A 40-year-old woman complained of severe episodic pain and sensitivity to cold exposure on the right thumb tip, left 4th and 5th finger tips for about 10 years. On physical examination there was erythematous to bluish tinge through the nail plates of the three fingers(Fig. 1). She denied a history of trauma to the digits and longterm medication of oral pills. Plain roentgenogram revealed a smooth concave erosion of the left 5th digital phalanx(Fig. 2). 7mm, 5mm and 4mm sized, soft encapsulated masses were excised from the three nail beds. Histopathologic examination revealed

Annals of Dermatology
Vol. 6, No. 2, July 1994

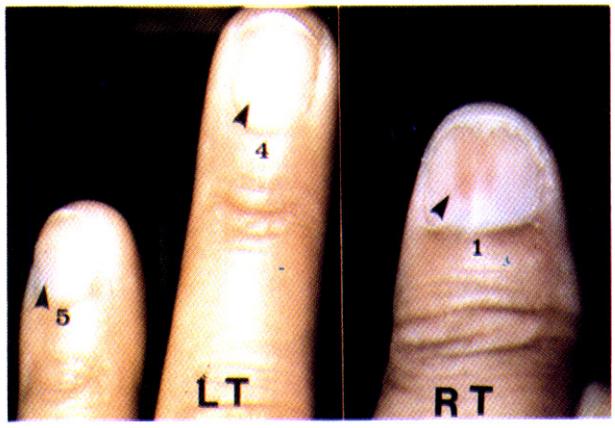

Fig. 1. Case 1. Erythematous to bluish tinge through the nail plates of the right thumb, left 4th and 5th fingers(▲).

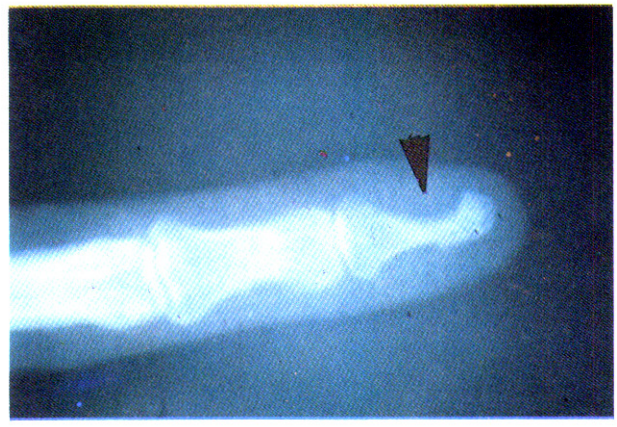

Fig. 2. Case 1. Plain X-ray film showing smooth concave erosiions along the unlar aspect of left 5th digital phalanx(▲).

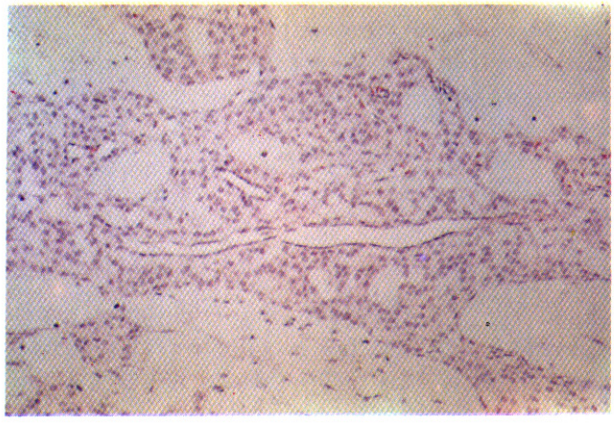

Fig. 3. Case 1. Multiple rows of glomus cells around the narrow vascular lumina(H & R stain, × 100).

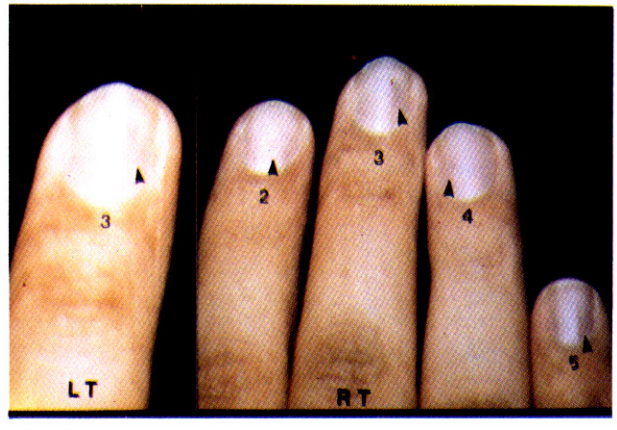

Fig. 4. Case 2. Faint erythematous or bluish tinge through the nail plates of fingers(▲)

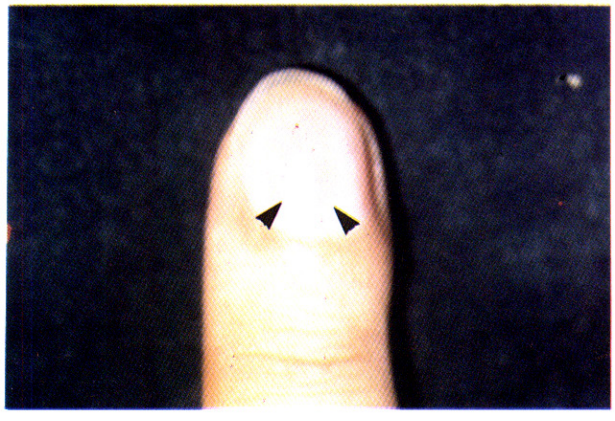

Fig. 5. Case 3. Slight bluish color change through the nail plate of the right 3rd finger(▲).

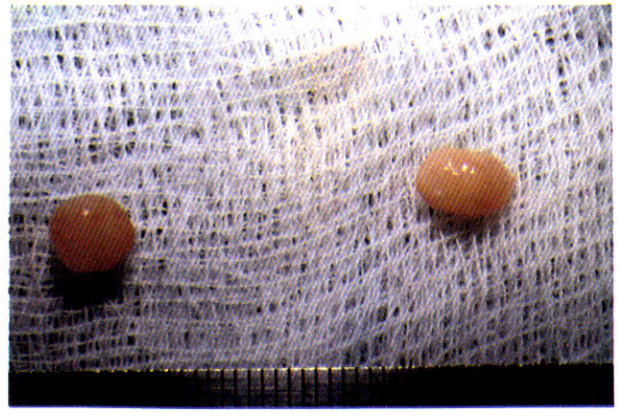

Fig. 6. Case 3. Two encapsulated tumor masses excised from the right 3rd finger tip.

the typical findings of solitary glomus tumors(Fig. 3).

Case 2. A 17-year-old girl presented with paroxysmal pain and tenderness on the right 2nd, 3rd, 4th, 5th finger tips, left 3rd finger tip and right great toe tip for about 3 years. Physical examination showed point tenderness and violaceous color change through the nail plates(Fig. 4) and also cafe-au-lait spots, axillary freckling and numerous soft pedunculated nodules on the whole body. She had experienced neurofibromatosis for the past 8 years. Plain roentgenogram showed well defined pressure erosion along the right 3rd digital phalanx. A diagnosis of glomus tumor was considered and excisional biopsy was done. Histopathologic findings of the six subungual masses were consistent with the solitary type of glomus tumor.

Case 3. A 25-year-old woman presented with tenderness and intense pain on cold exposure on the right 3rd finger tip for 8 years and a slight bluish color change was seen on physical examination(Fig. 5). There was no past history of trauma to the fingers. The radiographs failed to demonstrate any apparant bony abnormality. A diagnosis of possible glomus tumor was considered and an excisional biopsy was performed. On exploration of the nail bed, an unexpected similar mass was discovered on the nail matrix 0.4cm proximal to the nail bed mass. Both of the masses were excised(Fig. 6) and revealed histopathologic characteristics of the solitory type of glomus tumor.

DISCUSSION

Glomus tumors of the skin are generally classified as being solitary or multiple and the more common solitary type is usually tender and painful. The hand is known to be the most common location and 25 to 65% of glomus tumors in hands are found in subungual location[9]. Jablon et al[9] reported that the symptom triad of solitary glomus tumor of the hand was pain, tenderness and sensitivity to cold, and Schugart et al[10] mentioned about the characteristics of pain: a stimulus such as pressure or heat triggers a paroxysm of pain that not infrequently radiates away from the tumor and may be fleeting or may persist for some minutes. The clinical symptoms of our cases fitted with these characteristics of solitary glomus tumor. The severity of symptom did not seem to increase in proportion to the number and duration of the lesions(Table 1), which Schugart et al[10] already described.

Histologically our cases showed typical findings of solitary glomus tumor. The tumor was surrounded by a fibrous capsule and there were several narrow vascular lumina lined by a single layer of flattened, elongated endothelial cells. Peripheral to the endothelial cells were multiple layers of

Table 1. Summary of present cases

	Case 1	Case 2	Case 3
Location	subungual right thumb, left 4th and 5th fingers	subungual right 2nd, 3td, 4th 5th finger tips, left 3rd finger tip and right great toe	subungual right 3rd finger tip
Sex/Age	F/40	F/17	F/25
No. of Lesions	3 in 3 fingers	6 in 6 fingers	2 in 1 finger
Pain	yes	yes	yes
Tenderness	yes	yes	yes
Histopathologic findings	solitary type	solitary type	solitary type
Associated disease	none	neurofibromatosis	none
Duration	10 years	3 years	8 years

Table 2. Soliary type of glomus tumor developed in multiple sites reported in the literature

	Sank et al(1992)	Maxwell et al(1979)			Plewes(1941)
		Case 1	Case 2	Case 3	
Location	left 3rd finger tip, right 5th finger tip	left 4th finger tip 5 in 1 fingers	right 5th finger tip 2 in 1 finger	left 4th finger tip 4 in 1 finger	right 5th finger tip 4 in 1 finger
Sex/Age	F/22	M/44	F/31	F/28	M/16
No. of Lesions	2 in 2 fingers	5 in l finger	2 in 1 finger	4 in finger	4 in 1 finger
Pain	yes	yes	yes	yes	yes
Tenderness	yes	yes	yes	yes	yes
Histopathologic findings	solitary type	solitary type	solitary type	solitary type	solitary type
Associated disease	none	none	none	none	none
Duration	4 years, a few months	15 years	10 years	2 to 3 years	10 years
Auther's designation	bilateral solitary glomus tumors	multiple digital glomus tumors	multiple digital glomus tumors	multiple digital glomus tumors	multiple glomus tumors

glomus cells.

The glomus tumor is said to be associated with physical stimulus such as local trauma or blood estrogen concentration. Kennedy et al[11] reported cases accompanied by multiple endocrine neoplasia. Our second case had neurofibromatosis as well as glomus tumors. Kohout et al[12] reviewed 57 cases of glomus tumor and found two children had multiple neurofibromatosis as well. The relationship between glomus tumor and neurofibromatosis is not clear. Our supposition is that there is no relationship between the two diseases, because neurofibromatosis is a disease of the nerve sheath and glomus tumor basically originates from the vascular smooth muscle cell.

Gupta et al[13] reported multiple painful glomus tumors developed as five small nodules on the back, both arms and legs. Although their case included multiple painful tumors like ours, theirs was histologically the typical multiple type of glomus tumor and its distribution on the body was not the usual site of the solitary glomus timor. An interesting point in our cases is that their clinical and histopathologic findings are those of the solitary glomus tumor(Table 1), but they are multiple in number and this kind of lesion is very uncommon. Our cases are not believed to be the multiple type of glomus tumor because, in addition to their histopathologic findings and characteristic symptoms of the solitary glomus tumor, the literature shows that no multiple type has been found in subungual locations as all our cases were.

Our extensive search of the literature could not reveal any solitary glomus tumor developing in as many nail beds as our cases. Although Sank et al[6] reported bilateral solitary glomus tumors in only two digits of different hands, they developed about 4 years apart(Table 2). In our first two cases, the lesions developed almost at the same time and in more digits. The third case showed two nodules at one nail bed. Maxwell et al[7] and Plewes[8] also reported painful multiple glomus tumors at one digit and their cases were those of the solitary type rather than those of the multiple type in view of the clinical and histopathologic findings(Table 2). Our cases suggest that a patient should be examined more carefully in order to see if another glomus tumor is present besides the overt tumor. Also the third case suggests that the physician may consider that the pain may be caused not only by recurrence due to an incomplete excision but by the development of a second glomus tumor when the patient complains of pain on a previous excision site. Our cases show that the glomus tumors thought to be the solitary type can develop in multiple sites in two ways. One is

the pattern that tumors develop separately in each of multiple sites like our first two cases and the case of Sank et al[6] and the other is the pattern of our third case and the cases of Maxwell et al[7] and Plewes[8], which included glomus tumors developed in multiple sites of one digit. We now consider that these cases could be classified as variants of solitary type or another unspecified group of glomus tumor.

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