CASE REPORT

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The Importance of Preoperative Imaging Study on a Solitary Neurofibroma Originated from the Digital Nerve

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This case is about a rare type of a solitary neurofibroma that originated from the digital nerve between the proximal phalanx of a finger and the web space, which was first misdiagnosed as giant cell tumor, ganglionic cyst, or fibroma originating from the tendon before radiologic studies were done. The preoperative magnetic resonance imaging (MRI) showed a non-enhanced well-circumscribed mass and the digital nerve was deviated to the volar-medial side due to the mass effect. Since neurofibroma is difficult to differentiate from others by physical examination, crucial information such as the connection between the mass and the nerve or the deviation of the digital nerve can be obtained by MRI findings. And it is important to plan the surgery safely from this information.

Keywords: Digital nerve, Neurofibroma, Preoperative imaging

INTRODUCTION

Neurofibroma is one form of neurofibromatosis which can occur in multiple or in a solitary nodule¹. Multiple neurofibromatosis is closely related to von Recklinghausen's disease which can be easily diagnosed by familial history along with physical examination². On the other hand, solitary neurofibroma without any underlying disease is rare^{1,2}. This is a case of solitary neurofibroma originating from a digital

nerve located between third metacarpal bone and third proximal phalanx which was misdiagnosed as giant cell tumor, ganglion cyst, or fibroma originating from tendon prior to the histopathology result.

CASE REPORT

A 31-year-old female patient visited Seoul St. Mary's Hospital presenting an asymptomatic 2×1 cm sized mass located in

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radial aspect of third proximal interphalangeal (PIP) joint area which occurred 2 to 3 years ago. A protruding mass was detected without any sensory changes (Fig. 1). The mass was growing in size but no particular symptoms were detected. A magnetic resonance imaging (MRI) with enhancement was performed since physical examination provided only limited information. Preoperative MRI revealed a non-enhancing well demarcated mass yielding low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. The mass appeared to be originating from the digital nerve causing it to be shifted toward midline by a mass effect (Fig. 2). An operation was

Fig. 1. Preoperative photograph shows diffuse swelling (black arrow) of the middle finger, without overlying skin changes.

performed through a 1.5 cm size lateral incision in third radial PIP joint area (Fig. 3). The mass was identified under microscope assisted view and dissection was carried out along the radial digital nerve. The mass having connection to the nerve sheath was resected completely (Fig. 4).

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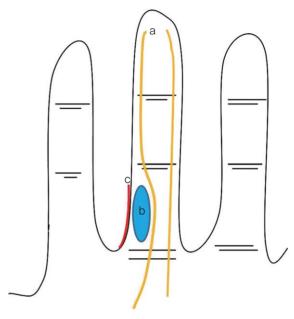


Fig. 3. A diagram of a solitary neurofibroma and a digital nerve shows the radial digital nerve(a) deviated to the midvolar side by the neurofibroma(b) and the incision design(c). Approaching towards the volar side of the finger could increase the chance of the digital nerve injuries.

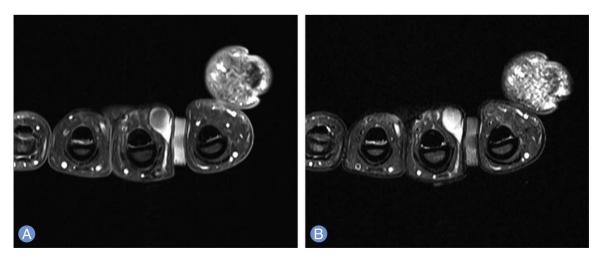


Fig. 2. (A) Neurofibroma which has a clear margin and connects to the radial digital nerve sheath with a low signal on T1-weighted images. **(B)** Neurofibroma with a non-enhanced high signal on T2-weighted images. Neurovascular bundle is deviated to the volar central portion.

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gated and wavy nuclei in a myxoid stroma were seen in hematoxylin and eosin staining (Fig. 5A). In immunochemistry, the positivity of S-100 protein revealed that the mass was Schwann cell origin and confirmed a neurofibroma (Fig. 5B). The negativity of CD68 differentiate it from schwannoma, which normally result in positivity for CD68. No sensory change was found postoperatively. No sign of recurrence was found 10 month follow-up.

DISCUSSION

Multiple neurofibromatosis type I is closely related to von Recklinghausen's disease accompanying change in skin

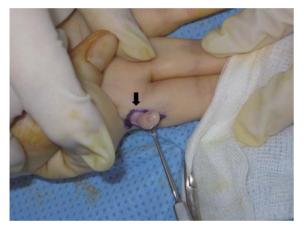


Fig. 4. Intraoperative photograph shows a neurofibroma attached to a digital nerve sheath by a stalk. Black arrow, radial digital nerve.

texture, café au lait spot, optic glioma, Lisch nodule^{2,3}. However, solitary neurofibroma has no particular symptoms except a growing mass, especially located in hands^{4,7}. Likewise the case presented, most cases are unnoticed because of its asymptomatic nature⁴.

Though it is difficult to diagnose solitary neurofibroma, MRI can provide preoperative diagnosis and relationship with digital nerve. A well demarcated mass having connection to digital nerve with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images strongly suggest solitary neurofibroma. Otherwise, giant cell tumors appears as solid masses, hypointense on both T1-and T2-weighted images⁸. About ganglion, this shows low to intermediate signals on T1-weighted images and high signals on T2-weighted images⁸. They may be uniloculated or multiloculated and contain proteinaceous synovial fluid⁸.

Sonography and computed tomography (CT) also could be helpful to differentiate diagnosis. Sonography can show in detail the tumor's site, size and echogenicity. Close contact of the tumor with tendon sheath, bone erosion, and internal vascularity can also be shown by sonography⁹. CT is superior to conventional radiography and tomography in outlining tumor extent especially its extra-osseous portion and its relationship to adjacent structures, as well as evaluation of cortical integrity and determination of tumor recurrence¹⁰.

Careful dissection under microscope was performed in

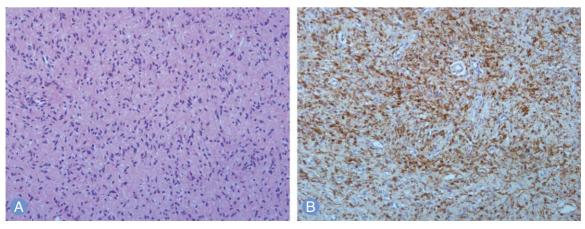


Fig. 5. (A) Irregular arranged spindle cells with enlongated and wavy nuclei in a myxoidstroma with thin collagen fibers scattered in between seen in H&E staining (\times 200). **(B)** Staining with S-100 protein helps detecting schwann cell origin tumors. Immunochemistry by S-100 shows deep brown colored cells (\times 200).

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order to maintain neural integrity in our case. If nerve damage should occur, prompt neurorrhaphy or nerve graft must be performed. In the case presented above, digital nerve shifted toward midline was confirmed preoperatively and dissection was performed from radial lateral incision distant from the nerve. In addition, the MRI revealed that the neurofibroma was connected to the nerve sheath in stalk rather than having a direct contact to the nerve. In the latter case, more meticulous dissection is required.

In case of a solitary neurofibroma occurring in hand, it is important to perform a preoperative imaging study in order to identify its relationship with digital nerve. If nerve damage is inevitable, sufficient warning should be given along with surgical planning for nerve grafting. Also, if an indistinctive mass apart from a common ganglion or a giant cell tumor is found, preoperative radiologic imaging studies are recommended in order to distinguish solitary neurofibroma.

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손가락 신경에서 기원한 단발성 신경섬유종에서 수술 전 영상 검사의 중요성

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본 증례는 드물게 손가락에서 지간으로 이어지는 부근의 손가락 신경에서 기원한 신경섬유종으로 영상학적 검사를 진행하기 전에는 거대세포종양 또는 결절종이나 힘줄에서 유래한 섬유종으로 오인했던 경우이다. 환자에게 수술 전 시행한 magnetic resonance imaging (MRI)에서 조영증강이 되지 않는 경계가 명확한 종괴가 관찰되었고 손가락신경이 종괴에 밀려 손바닥 쪽 및 안쪽으로 편위되어 있는 것을 확인하였다. 신경섬유종은 영상 검사를 시행하기 전까지 이학적 검사로 감별하기 쉽지가 않으며 신경집에서 유래하기 때문에 수술 전 MRI 등의 영상 확인을 통해 손가락 신경과의 관계, 손가락 신경의 위치 변위 등에 대한 정보를 미리 얻는 것이 수술을 진행하는데 많은 도움이 된다.

색인단어: 손가락 신경. 신경섬유종. 수술 전 영상 검사

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