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Subungual exostosis with post-operative recurrence followed by spontaneous regression

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Dear editor,

Subungual exostosis (SE) is a relatively uncommon osteocartilagenous tumor of the digital phalanges. Although post-operative recurrence sometimes develops, cases with spontaneous regression have not been reported. We report a case of SE with post-operative recurrence followed by subsequent spontaneous regression.

A 9-year-old girl presented with a 6-month history of a growing, painful nodule on the right hallux. Physical examination revealed a 10 mm nodule extending from beneath the nail plate of the hallux (Fig.1a). Radiographs revealed a calcified lesion which was continuous with the distal phalangeal cortex (Fig.1b). SE was suspected and the nodule was resected, including the base at the cortex, until cancellous bone was observed. Intraoperative radiographs confirmed optimal removal of the calcified lesion (Fig. 1c). Histology of the resected tissue revealed a normal trabecular bone and fibrocartilaginous tissue, consistent with SE (Fig. 1d). One month after the surgery, the nodule developed again and grew until 2 months after the initial surgery. Radiographs then demonstrated a calcified lesion at the same location, confirming recurrence of SE (Fig. 1e), and a proposed re-excision was refused by the patient and family. In spite of a lack of surgical intervention, however, the nodule gradually decreased in size and finally disappeared 8 months after the surgery. Radiographs also revealed that most of the

calcified lesion had spontaneously regressed (Fig. 1f).

The precise pathogenesis of SE remains unknown, but trauma and hereditary abnormalities have been suggested as possible inducing factors for SE. The recurrence of SE sometimes happens within a few months after surgery[1, 2]. In contrast, there have been no reports of SE with spontaneous regression, although only a few cases of exostosis with spontaneous regression in other bones (such as the humerus and radius) have been reported[3]. Since a fracture of the lesion or adjacent regions may be followed by regression of exostosis, previous reports pointed out that bone remodeling induced by trauma is one of the possible mechanisms[3]. Generally, the bone fracture repair process consists of 3 phases: inflammation, renewal, and remodeling[4]. Numerous cytokines and growth factors such as interleukin(IL)-1, IL-6, and transforming growth factor (TGF)-β are produced at the injury site in the inflammation phase, promoting the proliferation and differentiation of mesenchymal stem cells into osteoblasts which form callus woven bone in the renewal phase[4]. In the remodeling phase, osteoblasts and osteoclasts mediate the replacement of the woven bone with lamellar bone through renewing and resorptive actions by diverse factors. IL-1, IL-6 and TGF-β are also involved in the regulation of the remodeling phase through promotion of osteoclast formation[4, 5]. Therefore, although the exact mechanism remains unclear, we speculate

that some factors such as IL-1, IL-6 and TGF-β induced by the surgery may promote not only the formation of woven bone in the renewal phase but also the resorption of the bone in the remodeling phase, resulting in post-operative recurrence followed by spontaneous regression of SE in our case. Our case suggests that trauma, including surgeries, may have a dual function for induction and regression of SE.

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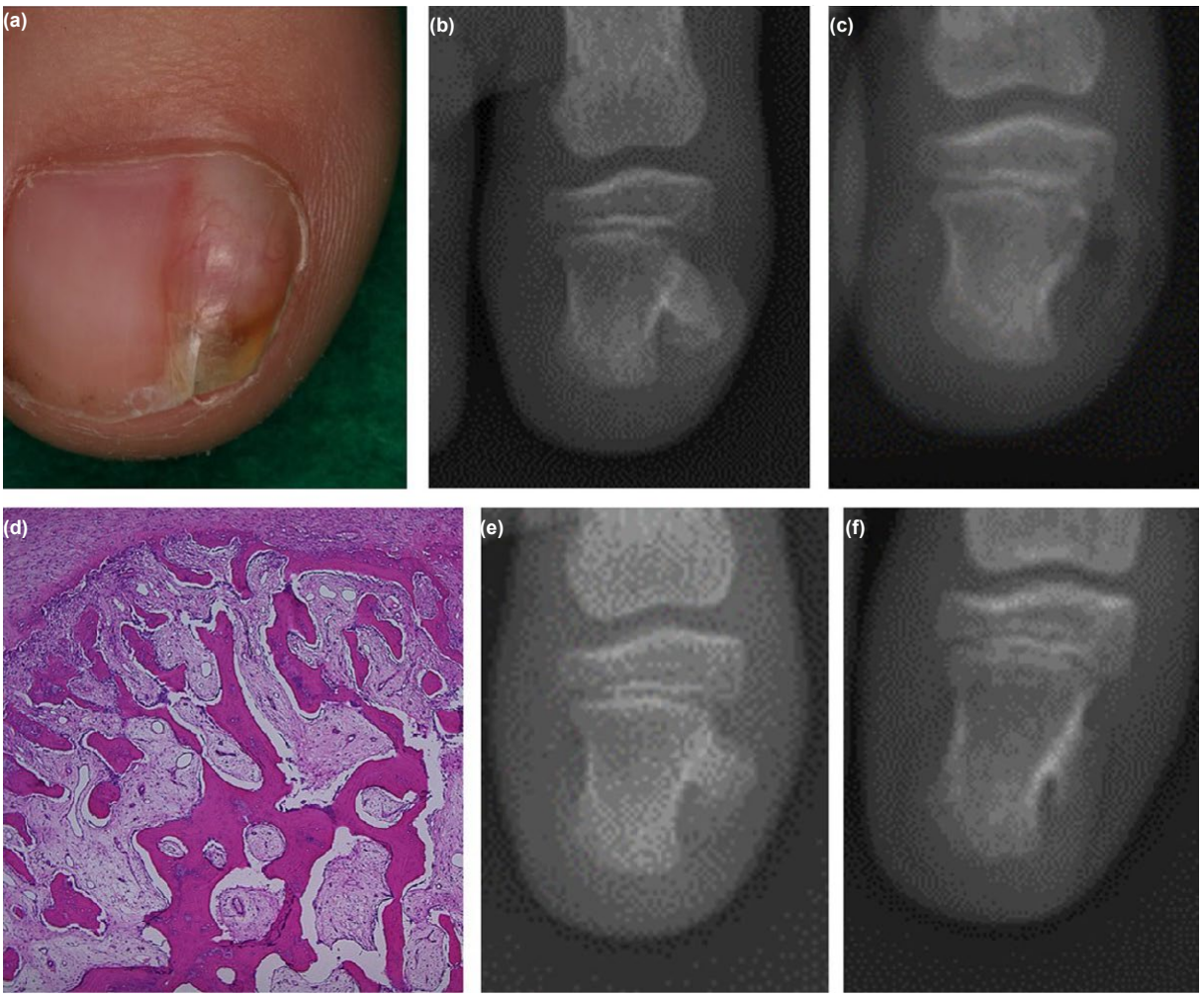

Figure 1. (a) A nodule beneath the nail plate on the right hallux. (b) The nodule showed a calcified lesion continuous with the distal phalangeal cortex. (c) Optimal removal of the lesion was confirmed by intraoperative radiography. (d) The lesion revealed a normal trabecular bone and fibrocartilaginous tissue (hematoxylin–eosin, original magnification 940). (e) A radiograph from 2 months after the surgery. A calcified lesion recurred at the same location. (f) A radiograph from 2 years after the surgery. Most of the recurrent calcified lesion spontaneously regressed.