CASE REPORT

WILEY

Subungual exostosis of the finger in an 8-year-old girl

Hao Li | Xinyu Qi | Xuejun Zhang

Department of Orthopaedics, Beijing Children's Hospital, Capital Medical University, Beijing, China

Correspondence

Xuejun Zhang, Department of Orthopaedics, Beijing Children's Hospital, Capital Medical University, Beijing 100045, China Email: zhang-x-j04@163.com

Received: 23 December, 2019 Accepted: 2 April, 2020

ABSTRACT

Introduction: Subungual exostosis (SE) is a relatively uncommon benign bone tumor that occurs in the distal phalanges of the toes or fingers.

Case presentation: An 8-year-old girl presented for treatment of an acquired mass on the distal right index finger. The patient was preoperatively diagnosed with a benign bone tumor. Surgical resection of the distal right index finger mass was performed under general anesthesia, and histological examination of the resected tissue supported a diagnosis of SE of the right index finger. The surgical outcome was good, with no surgical site infection. Throughout 15 months of follow-up, the patient was asymptomatic with no recurrence.

Conclusion: Surgical resection of a mass on the distal right index finger resulted in absence of recurrence during 15 months of follow-up. SE is a benign and uncommon lesion that is infrequently encountered by physicians. This infrequent occurrence may result in delays in diagnosis and treatment. Complete excision of the lesion and careful separation from underlying nail bed structures results in total resolution of the lesion, while providing the lowest risk of recurrence.

KEYWORDS

Subungual exostosis, Nail surgery, Hyperostosis, Nail tumors

INTRODUCTION

Subungual exostosis (SE) is a benign hyperosteogeny and relatively rare type of osteochondroma that is firmly attached to the distal phalanges of toes or fingers; this condition is most commonly observed during adolescence.^{1,2} The etiopathogenesis of SE is unclear, but may be associated with micro-trauma and infection.³ This benign lesion has also been regarded as a component of multiple hereditary exophytic osteochondroma.² Herein, we describe a patient who presented with SE on the right index finger, associated with repeated trauma to the nail bed.

DOI: 10.1002/ped4.12234

CASE REPORT

An 8-year-old girl presented with a slow growing painful swelling under the nail of the right index finger, which had been present for 2 years. She had no history of prior trauma or chronic infection. Physical examination revealed a firm, protuberant lesion of $0.8 \text{ cm} \times 0.5 \text{ cm}$ under the right index finger; no erythema or signs of an infection were observed (Figure 1A). Radiographic examination revealed a calcified projection on the distal end of the distal phalanx of the right index finger, which was continuous with the underlying normal bone (Figure 1B, C). Clinical and radiologic features supported a diagnosis of

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made. ©2020 Chinese Medical Association. *Pediatric Investigation* published by John Wiley & Sons Australia. Ltd on behalf of Futang Research Center of Pediatric

©2020 Chinese Medical Association. Pediatric Investigation published by John Wiley & Sons Australia, Ltd on behalf of Futang Research Center of Pediatric Development.

SE. Following surgical resection of the lesion (Figure 2A, B), histological examination revealed mature trabecular bone within the tumor, with a cartilaginous cap on the surface, as well as the formation of cartilage with reactive atypia. These findings were consistent with a diagnosis of osteocartilaginous exostosis (Figure 3). The benign tumor growth was completely removed by surgery and did not recur throughout 15 months of follow-up.



FIGURE 1 Clinical photos and radiographs of subungual exostosis in the right index finger of an 8-year-old girl. (A) Clinical photos, (B) posteroanterior and (C) lateral radiographs, of the lesion.



FIGURE 2 Intraoperative images of excision of subungual exostosis from the right index finger of an 8-year-old girl. (A) The lesion invaded the nail bed; thus, the nail bed could not be saved. Accordingly, total tumor resection was performed. (B) Osteochondral lesions were completely removed with sufficient margin.



FIGURE 3 Histopathological analysis (hematoxylin and eosin stain) revealed normal trabecular bone and fibrocartilage were overgrown; these findings were consistent with the diagnosis of subungual exostosis (×40).

DISCUSSION

SE is often encountered in adolescent patients.¹ The prevalence of SE is unclear, and it may be underestimated because of delays in diagnosis due to its unclear and inconsistent clinical manifestations.⁴ Most SE lesions are located on the large toe, although they may occasionally occur in other toes.⁵ Only a small proportion of SE lesions occur in the fingers.⁶ The most commonly affected site is the inner or medial aspect of the large toe.⁷

SE is a benign, slow-growing osteochondroma of the phalanges of toes or fingers. First described by Dupuytren in 1847, SE is the most common nail tumor of young adults, representing a benign bony proliferation of the distal phalanx with unknown etiology.^{7,8} The pathology diagnosed by podiatry specialists as SE does not comply with the epidemiologic, etiologic, clinical, and radiologic criteria of Dupuytren's SE. In accordance with the parameters of true exostosis (and therefore termed by some authors from other specialties as osteophyte of the distal phalanx), it should be considered as a different pathologic entity, and the surgical approach must also be different.⁹ SE typically occurs as a small, firm, solitary lesion, located deep to the free edge of the nail in children and young adults.⁴ Although chronic stimulation is regarded as the cause of fibrocartilaginous metaplasia, its pathogenesis remains unclear. Most clinicians presume that SE is a type of reactive metaplasia caused by microtrauma¹⁰; however, there is no conclusive evidence to support a single pathogenesis.¹¹ The proposed etiopathogenesis of SE includes chronic infections, trauma, hereditary abnormality, tumor, or activation of a cartilaginous cyst.¹² Some studies have shown that trauma is a major factor in the formation of SE, followed by acute and chronic inflammation leading to chondrometaplasia.¹³ Notably, our patient had no history of prior trauma or chronic infection.

SE typically occurs as a firm, fixed nodule with a hyperkeratotic surface at the distal end of the phalanx, under the nail plate (for example, as shown in Figure 1A). As the SE increases in size, the nail plate is lifted and detached from the underlying nail bed. The condition follows a chronic progressive course; pain and onychodystrophy are the most common complications.¹⁴ Because misdiagnosis and delayed diagnosis are common, appropriate treatment is often unavailable.¹⁵ Histologically, the cartilage cap of the exoskeleton is composed of fibrocartilage, while hyaline cartilage joins the osteochondroma trabecula and underlying cortical bone.¹⁶ Histology remains important for the diagnosis of suspected subungual lesions. Patients with a diagnosis of SE present with complaints of pain involving the nail and are diagnosed by radiographic evaluation. X-ray is the main diagnostic tool, which reveals excessive bone growth on the dorsal side of the distal phalanx (for example, as shown in Figure 1B, C); this bone growth

is composed of trabeculae. Pathological verification of diagnosis is important for follow-up. Dermoscopy may be a useful auxiliary tool to confirm the diagnosis of this benign nail condition, although X-ray examination and histopathology are necessary for definitive diagnosis.¹⁷ Dermoscopy enables differentiation from other nail conditions and can be used to guide patient treatment.¹⁸ The differential diagnosis of SE includes squamous cell carcinoma of the nail bed, verruca vulgaris, glomus tumor, subungual fibroma/fibrokeratoma, subungual epidermal inclusion cyst, pyogenic granuloma, achromic malignant melanoma, melanotic whitlow, osteogenic sarcoma, and enchondroma.¹³ The clinical manifestation of enchondroma is similar to that of SE; however, enchondroma is radiolucent and causes expansion of the phalanx itself.¹⁹ SE should be considered in the differential diagnosis of painful subungual tumoral lesions, particularly when a yellow spot is observed on the nail plate immediately above the subungual mass.²⁰

The effects of non-surgical treatment are limited because they do not occur with sufficient speed. The first choice of treatment is complete surgical resection. The main purpose of surgical treatment of SE is removal of exogenous bone to the normal bone tissue boundary (for example, as shown in Figure 2A, B), prevention of recurrence by protecting the finger matrix, and avoidance of nail deformity. Excision of the entire lesion, including the fibrocartilaginous cap, is crucial for prevention of local recurrence.²¹ Postoperative recurrence of SE has been reported, followed by spontaneous regression.²² Complete excision of the lesion and careful separation from underlying nail bed structures yield the lowest rates of recurrence and future complications.²³ In SE surgery, surgical excision of the mass without damaging the nail bed is important. The nail bed should be repaired after removal of the mass if it has been damaged during surgery. In our patient, there was no recurrence throughout 15 months of follow-up.

We have described a patient with SE of the right index finger, which has rarely been reported in the literature. SE is a rare benign disease, often associated with delayed diagnosis and treatment. X-ray examination can be used for the differential diagnosis of SE. Surgical removal is the main treatment modality; careful excision is needed to avoid damage to the nail bed and subsequent nail deformity. Thorough removal of pathological tissue is critical for prevention of recurrence.

CONSENT FOR PUBLICATION

Consent was obtained from the patient's guardians.

CONFLICT OF INTEREST

The authors have no conflicts of interest to disclose.

REFERENCES

- Hoehn JG, Coletta C. Subungual exostosis of the fingers. J Hand Surg Am. 1992;17:468-471.
- Mavrogenis AF, Papagelopoulos PJ, Soucacos PN. Skeletal osteochondromas revisited. Orthopedics. 2008;31:orthosupersite.com/view.asp?rID=32071.
- Vazquez-Flores H, Dominguez-Cherit J, Vega-Memije ME, Saez-De-Ocariz M. Subungual osteochondroma: Clinical and radiologic features and treatment. Dermatol Surg. 2004;30:1031-1034.
- DaCambra MP, Gupta SK, Ferri-de-Barros F. Subungual exostosis of the toes: A systematic review. Clin Orthop Relat Res. 2014;472:1251-1259.
- Evison G, Price CH. Subungual exostosis. Br J Radiol. 1966;39:451-455.
- Carroll RE, Chance JT, Inan Y. Subungual exostosis in the hand. J Hand Surg Br. 1992;17:569-574.
- 7. Russell JD, Nance K, Nunley JR, Maher IA. Subungual exostosis. Cutis. 2016;98:128-129.
- Aggarwal K, Gupta S, Jain VK, Mital A, Gupta S. Subungual exostosis. Indian J Dermatol Venereol Leprol. 2008;74:173-174.
- Perez-Palma L, Manzanares-Cespedes MC, de Veciana EG. Subungual exostosis: Systematic review and meta-analysis. J Am Podiatr Med Assoc. 2018;108:320-333.
- Storlazzi CT, Wozniak A, Panagopoulos I, Sciot R, Mandahl N, Mertens F, et al. Rearrangement of the *COL12A1* and *COL4A5* genes in subungual exostosis: molecular cytogenetic delineation of the tumorspecific translocation t(X;6)(q13-14;q22). Int J Cancer. 2006;118:1972-1976.
- Suga H, Mukouda M. Subungual exostosis: A review of 16 cases focusing on postoperative deformity of the nail. Ann Plast Surg. 2005;55:272-275.
- Guarneri C, Guarneri F, Risitano G, Lentini M, Vaccaro M. Solitary asymptomatic nodule of the great toe. Int J Dermatol. 2005;44:245-247.
- Ilyas W, Geskin L, Joseph AK, Seraly MP. Subungual exostosis of the third toe. J Am Acad Dermatol. 2001;45:S200-201.
- Das PC, Hassan S, Kumar P. Subungual exostosis Clinical, radiological, and histological findings. Indian Dermatol Online J. 2019;10:202-203.
- Smith LC, Funkhouser WK, Jr., Campion ER, Morrell DS. A firm nodule on the great toe of a 13-year-old boy. Pediatr Dermatol. 2009;26:747-748.
- Mertens F, Moller E, Mandahl N, Picci P, Perez-Atayde AR, Samson I, et al. The t(X;6) in subungual exostosis results in transcriptional deregulation of the gene for insulin receptor substrate 4. Int J Cancer. 2011;128:487-491.
- Piccolo V, Argenziano G, Alessandrini AM, Russo T, Starace M, Piraccini BM. Dermoscopy of subungual exostosis: A retrospective study of 10 patients. Dermatology. 2017;233:80-85.
- 18. Piccolo V, Russo T, Rezende LL, Argenziano G. Subungual

exostosis in an 8-year-old child: Clinical and dermoscopic description. An Bras Dermatol. 2019;94:233-235.

- Kumar P, Alam G. Subungual exostosis on index finger in a child. Indian J Dermatol Venereol Leprol. 2018;84:232-233.
- Demirdag HG, Tugrul Ayanoglu B, Akay BN. Dermoscopic features of subungual exostosis. Australas J Dermatol. 2019;60:e138-e141.
- 21. Wollina U, Baran R, Schonlebe J. Dystrophy of the great toenail by subungual exostosis and hyperostosis: Three case reports with different clinical presentations. Skin Appendage Disord. 2016;1:213-216.
- Nakamura Y, Maruyama H, Fujisawa Y, Inoue S, Okiyama N, Ishitsuka Y, et al. Subungual exostosis with postoperative recurrence followed by spontaneous regression. J Dermatol. 2019;46:e86-e87.
- Tchernev G, Grigorov Y, Philipov S, Chokoeva A, Wollina U, Lotti T, et al. Subungual exostosis in a young soccer player. Open Access Maced J Med Sci. 2017;6:52-54.

How to cite this article: Li H, Qi X, Zhang X. Subungual exostosis of the finger in an 8-year-old girl. Pediatr Investig. 2020;4:292-295. https://doi.org/10.1002/ped4.12234