

Does a Bone Deformity of the Distal Phalanx Undergo Remodeling after Removal of a Congenital Ectopic Nail?: A Case with Periodic Radiographic Follow-up

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Abstract

Background: Congenital ectopic nails are rare and are characterized by the presence of nail-like tissue mainly at the tip of a finger or toe.

Objective: Although the accompanying bone deformity might undergo remodeling after removal of the ectopic nail, it remains unknown whether complete bone remodeling can be eventually achieved, and whether such remodeling is necessary to improve the final cosmetic appearance of the concerned fingertip.

Methods and Results: Follow-up with periodic radiographic examination for 1 year after surgery in a child with congenital ectopic nail revealed no bone remodeling, despite the satisfactory cosmetic result.

Conclusion: We conclude that residual bone deformity does not affect the final appearance.

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Key words: ectopic nail, bone deformity, nail deformity, surgical treatment

Introduction

Only 26 cases of congenital ectopic nail have been reported, since Ohya first referred to this anomaly in 1931¹⁻²⁰ (**Table 1**). If the ectopic nail occurs at the finger tip, an M- or Y-shaped bone deformity of the distal phalanx is occasionally identified on radiography²⁻⁹. It has been suggested that an accompanying bone deformity might undergo remodeling after removal of the ectopic nail². Only Yamasaki, et al. have reported a case in child in which M-shaped bone deformity became shallow 4 weeks after removal of the ectopic nail². To clarify

whether bone remodeling can be achieved after the removal of an ectopic nail and whether there is any relation between postoperative cosmetic appearance of the concerned fingertip and bone remodeling. The patient was informed and consented that data concerning this case would be submitted for publication.

Case Report

A 3-year-old girl presented with a painless, hard keratotic projection, 2 mm wide and 3 mm long, on the tip of the left ring finger (**Fig. 1**). The projection had been present since birth just under the normal

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Table 1 All reported cases of ectopic nail, underlines indicate co-existed double fingernails.

	author	pub year	Age (y/o)	gender	location	Bone deformity
1	Ohya (1)	1931	21	F	Rt. thumb	
2	Kikuchi (13)	1978	40	M	Lt. V finger	
3			55	F	Lt. III finger	
4	Miura (19)	1978	7	F	Bil. V fingers & <u>Lt. IV finger</u>	+
5			?	M	Rt. V finger	
6	Aoki (14)	1983	71	M	Rt. II toe	
7			37	F	Lt. V finger	
8	Yamazaki (2)	1984	5	F	Lt. V finger	+
9	Katayama (18)	1984	9	M	Lt. V finger	
10	Higashi (3)	1984	13	M	Rt. V finger	+
11	Markison (11)	1988	31	F	Rt. III toe	
12	Aoki (16)	1991	35	M	Bil. V fingers	
13	Higashi (20)	1992	25	M	Rt. thumb	
14	Isago (4)	1995	16	M	Lt. V finger	+
15	Muraoka (12)	1996	4	M	<u>Rt. V finger</u> , Lt. V finger	
16	Shibata (5)	1996	28	M	Lt. V finger	+
17	Kuniyuki (6)	1996	26	F	Rt. V, Lt. IV finger	+
18	Iida (7)	1997	43	F	Bil. V finger	+
19	Tomita (15)	1997	23	F	Rt. V finger	
20	Hayashi (8)	1997	5	M	Rt. V finger	+
21			15	F	Rt. V finger	+
22	Kimura (17)	1997	0	M	Bil. V fingers	
23	Kamibayashi (9)	1998	6	F	Lt. V finger	+
24			4	M	Rt. V finger	+
25	Ena (10)	2003	34	M	Rt. sole	
26			27	M	Lt. heel	
27	This case	2005	3	F	Lt. IV finger	+

nail and had been trimmed by the patient's mother periodically because of its slow continuous growth. Neither active nor passive motions at any joint of that finger were not restricted. Radiography disclosed a Y-shaped bifurcation of the tip of the distal phalanx of the left ring finger, corresponding to the location of the projection (**Fig. 2**). No other anomalies were identified and the family history was not contributory.

Under general anesthesia, the projection regarded as a "small nail" was removed with the surrounding tissue. The surgical wound was approximated directly. Histologic examination revealed a keratogenous zone, possibly of the nail matrix, and nail tissue protruding through the normal horny layer of the epidermis, namely a small but complete nail matrix with normal nail plate (**Fig. 3**).

The postoperative course was uneventful, and no recurrence has been observed so far (**Fig. 4**).

Although the patient and her parents were satisfied with the cosmetic appearance of the fingertip, no diachronic bone remodeling of the bifurcated deformity in the distal phalanx had not been observed radiographically for more than 1 year after the removal of the ectopic nail (**Fig. 5a, b**).

Discussion

Twenty-four cases of congenital ectopic nail, including the present case, have been reported in Japan^{1-9,12-20}, and 3 cases have been reported in other countries^{10,11}. Because there is no logical epidemiologic explanation why most cases would occur in Japan, ectopic nail might be regarded as an unremarkable anomaly in other countries³. Although there are two main theories concerning the etiology of ectopic nail, namely the polydactylism theory and the embryonic aberration theory^{1,4,13,20}, a worldwide



Fig. 1 Preoperative view of the left ring finger with, small keratotic projection at the tip.



Fig. 2 Radiography disclosed a Y-shaped bifurcation of the tip of the distal phalanx.

consensus has not been achieved. Furthermore, whether double fingernail²¹ should be regarded as analogous to ectopic nail or the same anomaly remains controversial^{4,8,15}. Double fingernail is also rare and been reported as circumferential fingernail²², claw-like finger nail²³, calm nail deformity²⁴, and palmer nail syndrome²⁵. Recently, Tomita, et al. have hypothesized that two nail anomalies would have different appearances derived from the same embryologic pathogenesis¹⁵. Although

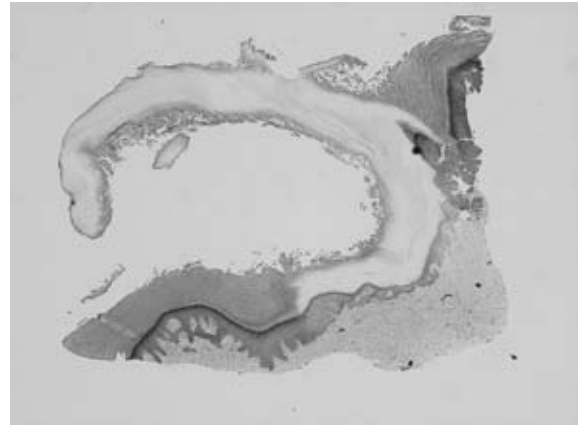


Fig. 3 A keratogenous zone, possibly of the nail matrix, and nail tissue protruding through the normal horny layer of the epidermis, namely a small but complete nail matrix with a normal nail plate. (Hematoxylin-eosin stain; original magnification, $\times 20$.)



Fig. 4 One year after surgery, no recurrence had been identified.

the two reports of ectopic nails co-existing with double fingernails may support a hypothesis^{12,19}, the difference in clinical characteristics between these two nail anomalies is quite apparent. In double fingernail, the corresponding finger is fusiform, and a rudimentary or well-formed nail can be observed on the volar aspect. Creases of the interphalangeal joints usually can not be traced. Both active and passive motions of each interphalangeal joint are highly disturbed. On the other hand, no disturbance of the joint motion has been reported with ectopic nail.

If an ectopic nail occurs at the fingertip, an M- or

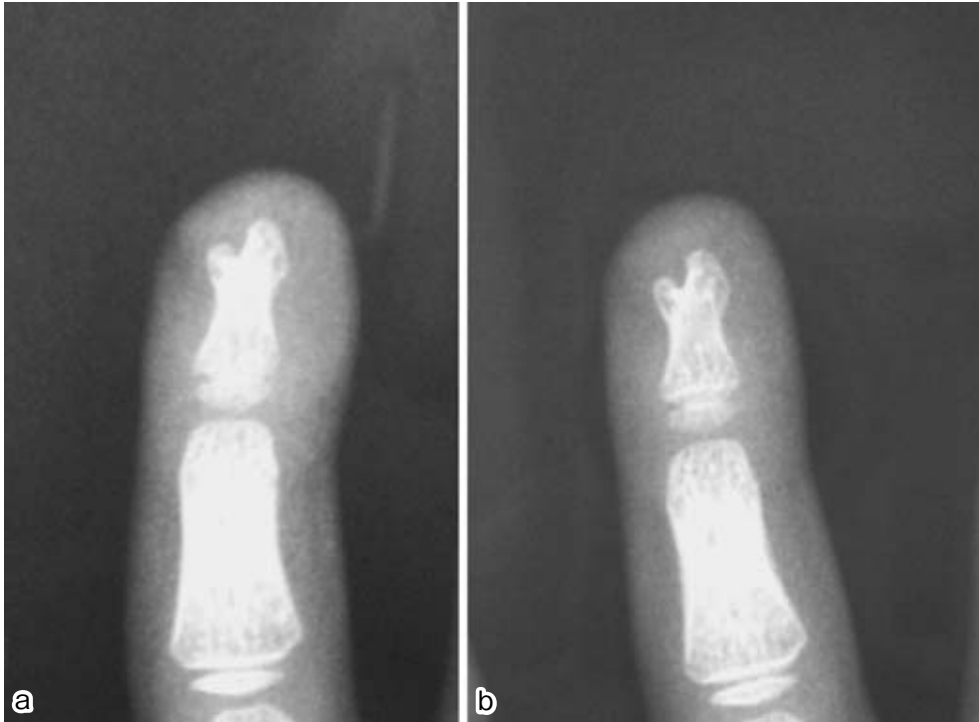


Fig. 5 Diachronic bone remodeling in the distal phalanx can not be identified after removal of the ectopic nail. **a:** 6 months after removal, **b:** 1 year after removal.

Y-shaped bone deformity of the distal phalanx is occasionally identified on radiography²⁻⁹. We clarified that even if a residual bone deformity does not resolve after the removal of the ectopic nail, it would not affect final cosmetic appearance of the concerned fingertip. Although some methods of local flap reconstructions after the removal of ectopic nails have been introduced for cosmetic reasons and for decreasing postoperative pain^{45,17}, simple surgical removal of an aberrant nail tissue and direct closure can generally provide satisfying results in childhood.

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