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Original Article

Exostosis in the Hand: Case Series and Literature Review
手部外生骨疣：系列病例報告與文獻回顧

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ABSTRACT

Introduction: This study reviews the exostosis in hand in our locality and compares those in the literature.
Methods: A retrospective study was conducted by reviewing the cases collected from 2000 to 2010. Literatures were also reviewed to look into the current concept of exostosis in the hand.
Results: There were nine patients with a total of 11 lesions of exostosis in the hand. Two cases were parosteal osteochondroma (Nora lesion). The male to female ratio was 4:5. The average presented age at the first consultation was 49.7 years of age. The most common site was distal phalanx. Six lesions had incorrect preoperative diagnoses by clinical assessment and standard two-view radiographs of the hand. Three of the six lesions were corrected after intraoperative Xi-scan screening.
Conclusion: The clinical picture of exostosis in the hand in our locality seems to be different from that of Western countries. Lesions of the hand were difficult to be visualized in the radiographs due to overlapping of bones. Xi-scan may help in making the diagnosis.

中 文 摘 要

簡介：本研究回顧手部外生骨疣的病例，並與文獻記載作出比較。
方法 ： 我們做了一個回顧性的研究，收集了本醫院在2000至2010年間處理的手部外生骨疣的病例。並且翻閱了文獻，從而了解現時醫學界對手部外生骨疣的概念。
結果：共9位病人的手部發現有11個外生骨疣的病例，當中兩個是骨膜外骨軟骨瘤;。男女的比例為4比5。首次求診的平均年齡為49.7歲。最常見的部位是在末指節骨。
經過臨床和手部標準X光評估，有6個錯誤的術前診斷。其中三個診斷可在手術中Xi-掃描檢查後得到更正。
結論 ： 本地區的手部外生骨疣在之臨床情況似乎與西方國家不同。由於骨骼在X光片中重疊，手部的病變有時難以看清。Xi-掃描可能有助其診斷。

Introduction

Exostosis in the hand *per se* is quite rare. However, it is the second most common osseous tumor of hand after chondroma.[1] There were only very few studies describing the clinical features in our locality. This study reviews the cases of exostosis in hand and the literature so as to update our knowledge of the condition.

Materials and methods

A retrospective study was conducted by reviewing the cases of exostosis managed in our centre from 2000 to 2010. The patients with clinical diagnosis of 'exostosis' were recruited. There were 55 cases in total. Eight of them had hereditary multiple exostoses with a positive family history. Among these eight patients, none of their lesions involved the hand. There were nine patients with 11 lesions of exostosis in the hand found. All of them had histologic proof.

Results

All of the nine cases of exostosis in the hand were of a solitary form. Among these nine patients, two had two solitary lesions at different sites over the hand. The clinical information of the nine patients is summarized in Table 1.

The male to female ratio was 4:5. The average age at the first consultation was 49.7 years of age with a mean duration of occurrence of 5 years. The mean diameter of lesion was 1.1 cm.

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Table 1
Summary of exostosis in the hands

Sex	Age	Number of lesion	Location	Diameter (cm)	Time lapse to the first consultation (years)	Symptoms	Physical examination	Severity of pain	Cosmetic problem	Functional impairment	Correct diagnosis before surgery	Preop diagnosis	Diagnosis changed after intra-op xi-scan	Histology
M	80	1	Right thumb metacarpal	3	1	2 + 3	3	1	2	1	Yes	Osteochondroma	No	Nora lesion
F	57	1	Right little finger proximal phalanx	1.5	8	1 + 2	1 + 2 + 3	2	2	1	No	Extra digit	Yes	Osteochondroma
M	41	1	Left thumb distal phalanx	1	>10	2 + 3	1 + 4	2	2	1	Yes	Osteochondroma	No	Osteochondroma
M	48	1	Right middle finger proximal phalanx	1	20	2	3	1	2	1	Yes	Osteochondroma	No	Nora lesion
M	58	1	Right thumb distal phalanx	1	1	2 + 3	1 + 3	1	2	1	Yes	Osteochondroma	No	Osteochondroma
F	64	1	Right hamate	1	1	1 + 2 + 3	1 + 3	2	1	2	Yes	Osteochondroma	No	Osteochondroma
F	54	1	Left middle finger distal phalanx	0.5	2.5	1 + 2 + 3	1 + 3	2	2	1	No	Osteophyte	No	Osteochondroma
F	17	2	Right thumb distal phalanx	0.5	0.5	1 + 2	1 + 3	2	1	2	No	Bony mass	Yes	Osteochondroma
			Right index finger distal phalanx	0.5								bony mass	Yes	osteochondroma
F	28	2	Left capitate	1	1	2	3	1	2	1	No	Ganglion	No	Osteochondroma
			Right thumb distal phalanx	1								ganglion	No	osteochondroma

Symptoms: 1 = pain; 2 = swelling; 3 = growing mass.
Physical examination: 1 = tenderness; 2 = numbness/Tinel positive; 3 = swelling; 4 = deformity.
Severity of pain, cosmetic problem, functional impairment: 1 = no; 2 = mild; 3 = moderate; 4 = severe.

Eight lesions were on the right hand and three were on the left. The most common site was the distal phalanx. The common symptoms and signs included a painless or painful swelling, a growing mass, numbness, and deformity of the finger. The lesions usually caused mild to moderate degree of cosmetic problems and functional impairment. There were two cases of parosteal osteochondroma (i.e., Nora lesion). Both of them showed the characteristic of incontinuity of the cortex and spongiosa of the underlying major bone (Figure 1). No subungual exostosis was found.

All nine patients had no family history of exostosis. One of them had a history of cut injury and one had previous surgery for release skin contracture at the same site. None of them had history of radiation exposure.

The clinical diagnosis of exostosis was made after clinical assessment and investigation with standard two-view radiographs of the hand. One patient was further worked up by blood tests and a computed tomography (CT) scan was performed as her radiographs showed bony erosion by the lesion. Six out of 11 lesions in four patients had incorrect preoperative diagnoses. After screening by the intraoperative Xi-scan, three out of the six lesions were amended as 'exostosis.' The remaining three misdiagnosed lesions were labeled as ganglions and osteophytes until histologic confirmation.

All of the nine patients had received marginal excisional biopsy of the lesions. Two patients had recurrence at 12 and 16 months after excision, respectively. However, re-exploration was not done in both cases.

Figure 1. The radiograph of patient with parosteal osteochondroma (Nora lesion) over proximal phalanx of the index finger.

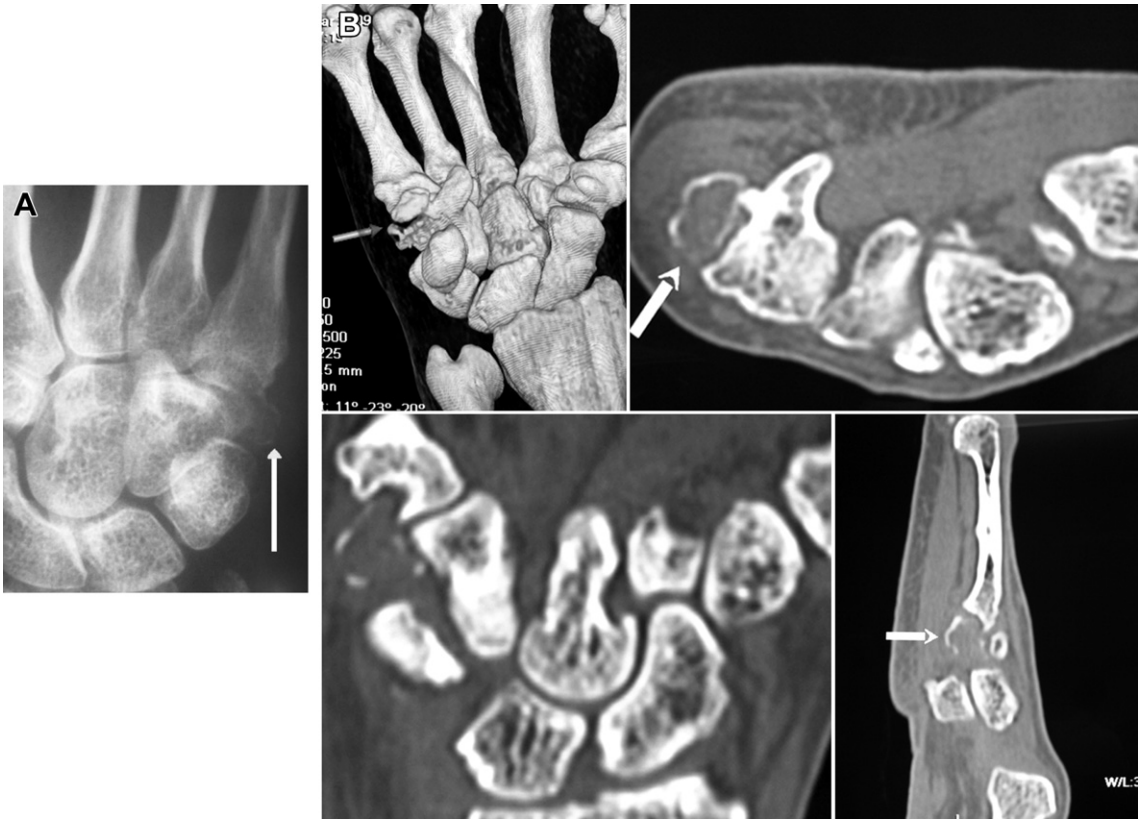

Figure 2. (A) The radiographs showed a bony mass ulnar to the base of the fifth metacarpal bone with erosion of surrounding bone; (B) computed tomography scan showing the lesion that indents and erodes into the base of the fifth metacarpal.

Exostosis is a benign lesion. Sometimes, it would present atypically, resembling an aggressive lesion. In our series, a 64-year-old woman presented with a painful swelling over the base of the right fifth metacarpal bone for more than 1 year. There was no limitation of joint movement or neurovascular problem. The radiographs showed a bony mass ulnar to base of the fifth metacarpal bone with erosion of the carpal-metacarpal joint, the base of the fifth metacarpal bone, and ulnar distal portion of the hamate (Figure 2A). Blood test showed normal while cell count (WCC) and alkaline phosphatase (ALP) levels. Erythrocyte sedimentation rate (ESR) was 17. CT scan was done. It showed a hypodense lesion 1 × 0.75 cm with incomplete hyperdense rim seen abutting the medial aspect of hamate hook. The lesion indented and eroded into the base of the fifth metacarpal distally (Figure 2B). Intraoperatively, there was a mass with cartilage cap abutting to base of the fifth metacarpal. Its stalk was communicating with the trabeculars of the hamate on the ulnar side. Excision of the mass exposed the marrow of hamate. The pathology of the mass was found to be an exostosis.

Lesions of the hand may be difficult to visualize due to the overlapping of other bones. Figure 3 shows the radiographs of the 57-year-old woman with the preoperative diagnosis of 'extra digit' that was changed to 'exostosis' after intraoperative Xi-scan. A 17-year-old girl who had solitary bony mass over the distal phalanx and her right thumb and right index finger. She had Apert syndrome with had multiple corrective surgeries done of her hands in childhood. Her lesions are not well shown in the preoperative two-view radiographs. However, the lesions could be seen clearly by Xi-scan (Figure 4) screening using special views.

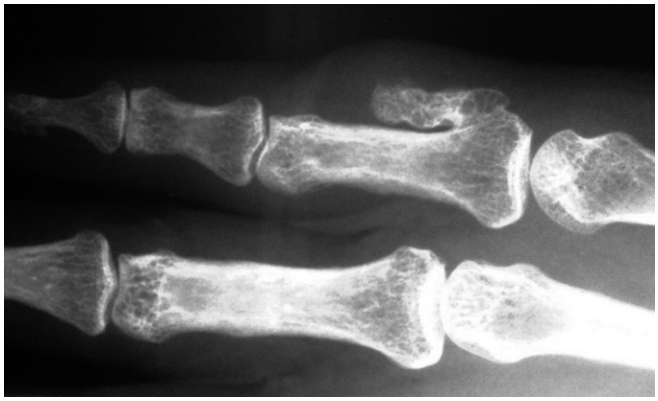

Figure 3. The radiograph of a 57-year-old woman with the preoperative diagnosis of extra digit.

Discussion and conclusion

Exostosis is the formation of new bone on the surface of a bone. It usually points away from the adjacent joint. One of the most common types is osteochondroma. Osteochondroma is a cartilage-capped bony projection on the external surface of a bone. It composes of cortical and medullary bone with overlying hyaline cartilage cap, and it is characterized by marrow and cortical continuity with the underlying parent bone. Osteochondroma commonly develops in bones during growth and increases in size throughout the childhood. After skeletal maturity, it usually has no further growth. The presentation is usually around 10 to 30 years of age. More than 60% are male patients.[1,2] Majority of cases presents as a painless swelling in metaphyseal region of long bones of hand

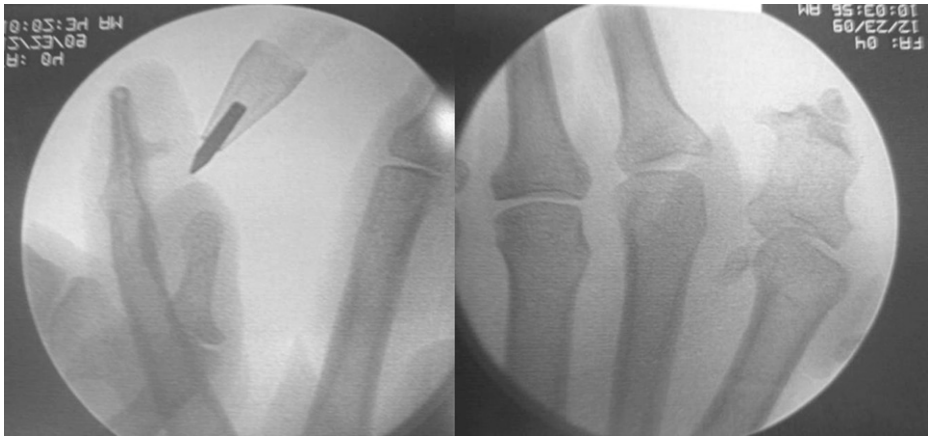

Figure 4. The lesions over index finger and thumb could be visualized clearly by Xi-scan at special views.

usually in the distal part of metacarpals and in the proximal part of proximal phalanx. Cases over the carpal bones were also reported, including the scaphoid and hamate.[3–5] The clinical features include asymptomatic painless masses; impingement causing irritation to muscle, nerves, ligaments, or tendons; limitation of joints movements; deformity resulting in cosmetic and functional impairment; and rarely pseudoaneurysm. Osteocartilaginous loose bodies or nodules of synovial chondromatosis may be found in the bursa nearby, which are called exostosis bursata. Malignant transformation of exostosis had been reported, although they are extremely rare.[6] The treatment for exostosis is mainly conservative since the risk of malignant transformation is less than 1%.[2] The indication for operative treatment includes pain from impingement or fractures, blocking of joint motion, cosmesis or severe deformity, and suspicion of malignant transformation. Surgical removal of the tumour should be flush to the bony surface, together with the whole cartilage cap. The recurrence rate was reported to be 2%.[1] Failure to remove the entire cartilaginous cap or the underlying periosteum and missing the diagnosis of a low-grade chondrosarcoma are the common causes of recurrence. Second surgery may be indicated in symptomatic recurrence and suspicion of malignancy.

According to the Western literature, osteochondroma is a disease of young patients.[1,2] However, the most common age group of patients with osteochondroma in the hand in our study were those of middle age. The most common site of osteochondroma in the hand among our patients was the distal phalanx, which is in contrast to the literature.

Subungual exostosis is a subtype of exostosis at the distal phalanx, and it was initially described by Dupuytren in 1847.[1] Subungual exostosis shares some radiographic features with an osteochondroma but it represents a distinct entity pathologically. The pathogenesis of subungual exostosis is believed to be chronic and repetitive trauma and infection. Proliferating fibroblasts are presumably induced by repeated trauma. Cartilage metaplasia then develops, which progresses to mature ossification. Classically, it arises from the dorsal or dorsomedial aspect of the distal phalanx, with a variable relationship of the nail bed. Majority of the lesion involves toes, which accounts for 80%–90%. The common presentations involve pain and swelling. Ulceration, infection, and nail deformity are frequent. The radiologic features comprise of outgrowth of trabeculated bone projecting from the distal portion of the distal phalanx. Cortex of the underlying bone does not flare out into the cortex of the subungual exostosis. There is no continuity of the lesion with underlying cortex and medullary canal, which differentiate it from osteochondroma. Histologically, spindle cells are not seen in classical exostosis but in subungual exostosis. Spindle cells proliferate to cartilage, and then to bony trabeculae through gradual maturation. Besides, the cartilage cap in subungual exostosis consists of fibrous cartilage rather than hyaline cartilage in osteochondroma. There was no subungual exostosis found in our study.

Parosteal osteochondromatous, also called Nora lesion, is another subtype of exostosis. It is a condition similar to subungual exostosis but occurs away from the nail bed.[7] It is more common in hands than feet. Patients usually present with painless swelling. In the radiographs, it appears as a well-demarcated mass of heterotopic calcification or ossification from the cortical surface of the affected bone. Similar to subungual exostosis, cortex and spongiosa of the lesion do not continue with the bone. Histologically, the cartilage can be a cap or be in lobules separated by dense fibrous tissue, which is usually hypercellular. The bone component is more irregular then osteochondroma. Spindle cells are arranged loosely between the trabeculae. It has a higher recurrence rate than osteochondroma. Cases of malignant transformation have never been reported. There were two cases of parosteal osteochondroma, i.e., Nora lesion, in our series. This lesion is not uncommon.

Lesions in the hand are sometimes difficult to be visualized in the radiographs due to overlapping of bones. Conventional two-view radiographs of the hand may be inadequate in making the diagnosis and the Xi-scan screening could help in some cases.

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