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Research Paper

Evolving Treatment Modality of Hand Enchondroma in a Local Hospital: From Autograft to Artificial Bone Substitutes

— 所地區性醫院治療手部內生軟骨瘤- 由自體骨移植術到使用人工合成骨的發展



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ABSTRACT

Background/Purpose: Curettage followed by the application of iliac crest autograft was the traditional treatment of hand enchondroma in Princess Margaret Hospital, Hong Kong.

Methods: We reviewed the results of 13 patients with hand enchondroma who were operated on in the past 15 years (1999–2013). Eight patients (1999–2009) received iliac crest autograft after curettage, whereas the other five patients (2009–2013) received artificial bone substitutes.

Results: Both groups of patients had good functional outcome and bone graft incorporation. There was no recurrence. One patient in the autograft group had mild residual finger stiffness. One patient receiving artificial bone substitutes had a gouty attack, early wound infection, and finger stiffness. The use of artificial bone substitutes eliminated donor site morbidities, decreased operation time and, hospital stay. They took a longer time for radiological incorporation than autograft, but it did not translate into adverse clinical effects.

Conclusion: Artificial bone substitute is a good alternative to iliac crest autograft in the treatment of hand enchondroma.

中文摘要

在香港瑪嘉烈醫院，治療手部內生軟骨瘤的傳統方法是刮除術和自體髌骨移植手術。我們回顧了最近15年(1999–2013)手部內生軟骨瘤的手術成效。由1999至2009年，8位病人接受了自體髌骨移植手術。由2009至2013年，5位病人接受了人工合成骨移植手術。兩組病人的手部功能回復理想，移植骨成功癒合，沒有復發的病例。一位接受自體髌骨移植手術的病人出現輕微手指僵硬的情況，而另一位接受人工合成骨移植手術的病人痛風症病發，出現傷口感染和手指僵硬的情況。使用人工合成骨可以避免髌骨傷口出現併發症，減少手術和住院時間。人工合成骨需要較長的時間癒合，但沒有帶來負面的臨床影響。使用人工合成骨來治療手部內生軟骨瘤是除了自體髌骨移植術以外的好選擇。

Introduction

Enchondromas are the most common benign bone tumours of the hand (35–65%).¹ They are islands of cartilage that persist in the bone after endochondral ossification. They can be seen at any age and in any bone preformed in cartilage, but are the most commonly found in the hands and feet. Enchondromas can be solitary or multiple. Multiple enchondromatosis is found in Ollier's disease or Maffucci's syndrome. Malignant transformation of a solitary enchondroma into chondrosarcoma is rare, but the risk increases to

25% in patients with Ollier's disease and 100% in those with Maffucci's syndrome.²

Patients with enchondromas may present with local pain and swelling, pathological fracture, or found incidentally on radiographs. Classic radiographs show a well-defined, central osteolytic lesion in the metaphyseal–diaphyseal junction. There may be expansion of the lesion with cortical thinning. They are most commonly found in the proximal phalanx and distal metacarpal bones of the hand.¹

Treatment of enchondromas aims to provide a histological diagnosis, maintain bony stability, prevent future fracture, and eliminate symptoms if any. Curettage is the treatment of choice.

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Some authors believe that a simple curettage is already enough for small defects and good bony stability because new bone formation is induced at the curettage site.^{1,3} However, the most common method used is to augment the bone defect with bone graft. The standard choice of bone graft was iliac crest autograft. In recent years, the appearance of commercially-packed and readily available artificial bone substitutes in the market expanded our armamentarium. Surgeons utilized different forms of artificial bone substitutes (e.g., demineralized bone matrix,⁴ calcium phosphate cement,⁵ and calcium sulfate pellets⁶). These bone substitutes provided good bone incorporation and functional outcome, while eliminating potential donor site complications associated with autograft.

Our hospital has started using artificial bone substitutes in the treatment of enchondroma since 2009. Up to then, iliac crest autograft was the standard source of bone graft. We reviewed the results of patients treated with both modalities.

Patients and methods

Between 1999 and 2013, 13 patients received operations for enchondroma of the hand. Seven patients were male and six were female. The average age of patient was 40 years old (range, 4–67 years old). There were 14 enchondromas in total. One patient had two enchondromas of the same hand (proximal phalanx of index finger and little finger) which were operated by two teams of surgeons simultaneously. All were sporadic cases. No case of Ollier's disease or Maffucci's syndrome was identified.

Six out of 13 patients (46%) presented with pain and swelling. Six other patients (46%) presented with pathological fracture preceded by a trivial injury. All were treated with splintage until the fracture healed before curettage and bone grafting were carried out. The average duration of splintage was 16.5 weeks (range, 10–21 weeks). In one patient (8%), the diagnosis was an incidental finding on a radiograph performed for another reason.

The most common site of occurrence of enchondroma was the proximal phalanx (10 out of 14 enchondromas; 72%). Two enchondromas occurred in the metacarpus (14%), one was found in the distal phalanx (7%), and one in the middle phalanx (7%). The order of location was similar to that quoted in the literature.

All 13 operations were performed under general anaesthesia. Dorsal approach was adopted and the extensor tendon was retracted. After adequate exposure, a cortical window was created on the bone with a burr (Figure 1). Thorough curettage was performed to evacuate the tumour (Figure 2), followed by water irrigation. The bone void was then filled with iliac crest autograft (Figure 3) or artificial bone substitutes (Figures 4 and 5). The cortical window was finally sutured back.

From 1999 to 2009, eight patients received autograft from the iliac crest (7 out of 8 patients, including the patient with 2 enchondromas) or the distal radius metaphysis (1 patient, 4 years old). In case of small enchondromas (2 out of 8 patients), a trephine biopsy needle was used to obtain iliac crest or distal radius bone graft. However, open harvest of iliac crest bone graft was done (6 out of 8 patients) for more sizeable lesions. The approximate volume of autograft used ranged from 0.5 mL to 4 mL. From 2009 to 2013, the other five patients received artificial bone substitutes, three of them received demineralized bone matrix putty (DBX Putty; Musculoskeletal Transplant Foundation and Synthes, Paoli, PA, USA), one patient received Norian SRS Bone Void Filler (Synthes), and one patient received ChronOS granules (Synthes).

All tumours were sent for histological examination and confirmed to be enchondroma. They were lobulated lesions consisting of clusters of chondrocytes with small and round nuclei. Myxoid changes, which can be seen in small bone chondroma, are noted. No chondrosarcoma was identified.

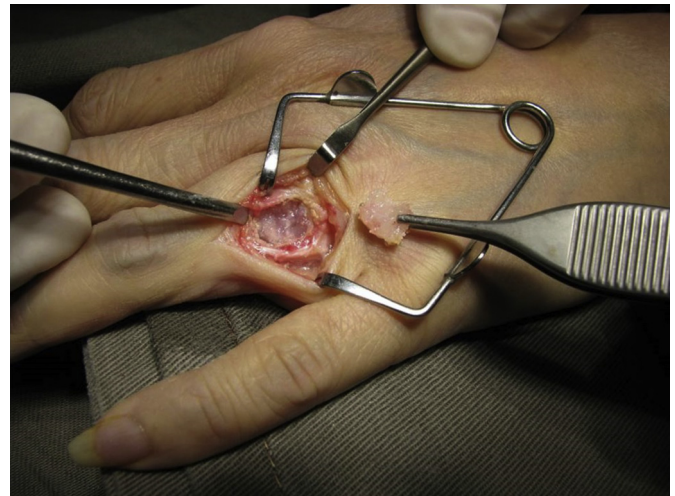


Figure 1. Exposure of the enchondroma after the cortical window was removed.



Figure 2. Curettage and evacuation of the enchondroma.



Figure 3. Application of an iliac crest autograft.



Figure 4. Application of Synthes DBX® putty.



Figure 5. Application of Synthes ChronOS granules.

Postoperatively, patients who presented initially with a pathological fracture and those who received artificial bone substitutes were put on splints for 4–6 weeks. More vigorous mobilization exercise was started after the course of splintage.

All patients were followed up in the Orthopaedics & Traumatology Specialist Out-patient Clinic of Princess Margaret Hospital to determine results and complications. Patients were asked to complete the Chinese *QuickDASH* questionnaire⁶ to determine their functional outcome. It consisted of 11 questions regarding activities of daily living and severity of symptoms, four optional questions regarding abilities of work, four optional questions regarding sports or music. A score of 1–5 was given to each question. Higher scores represented increasing difficulty in performing certain activities. The final score was expressed in percentages, representing the degree of functional disability. Serial X-rays were taken at the clinic to determine the radiographic outcome.

Results

The average operating time of the autograft group was 76 minutes (range, 47–155 minutes). The patient with two enchondromas was counted as two separate procedures, and the operating

time was 150 minutes and 155 minutes by two teams of surgeons respectively. Curettage followed by the application of artificial bone substitutes required an average of 52 minutes (range, 35–71 minutes).

The average duration of hospital stay for patients receiving iliac crest or distal radius autograft was 2.75 days (range, 2–5 days). For patients receiving artificial bone substitutes, the average duration of hospitalization was 1.3 day (range, 0.5–2 days).

All patients attended regular follow-up in the Specialist Orthopaedic Clinic after the operation. The average duration of follow-up of the autograft group was 25 months (range, 12–44 months). Because artificial bone substitutes were introduced in our hospital in more recent years (from 2009 to 2013), their follow-up duration was relatively shorter than the autograft group, averaging 17 months (range, 4–42 months).

In general, patients treated with either modality had good functional outcome. The average Chinese *quickDASH* score⁷ of the autograft group was 2.4. All patients receiving autograft had no residual pain over the digit, seven out of eight (87.5%) had full range of motion, and all of them had full hand grip power. Only one of them who presented with a pathological fracture of the proximal phalanx initially and required an external fixator (Synthes Mini External Fixator) after bone grafting had a flexion contracture of 10° of the proximal interphalangeal joint. None of them had wound infection or residual pain of the iliac crest wound.

Patients receiving artificial bone substitutes had similar functional results. Their average Chinese *quickDASH* score⁷ was 2.75. All patients had no residual pain. Four out of five patients (80%) achieved full range of motion and full hand grip power. One of them was complicated with gouty attack, early wound infection, and stiffness of fingers. He had known gouty arthritis and multiple gouty tophi over the operated digit. There were preexisting osteoarthritic changes of the finger joints. The surgical trauma induced a gouty attack which led to wound dehiscence, drainage of tophaceous material, and finally resulted in wound infection. The wound eventually healed up at postop 8 weeks with antibiotics and regular dressing.



Figure 6. Images of preoperative and 3 months postiliac crest bone graft application: bone graft is well incorporated.



Figure 7. Synthes DBX[®] putty incorporated after 4 months.

The progress of bone graft incorporation was determined on serial radiographs. Autografts were consolidated in a mean period of 5 weeks (range, 4–6 weeks; Figure 6). Artificial bone substitutes were incorporated in a mean period of 19 weeks (range, 12–33 weeks; Figure 7). No case of nonunion or recurrence was detected clinically or radiologically upon the latest follow-up.

Discussion

The treatment of enchondroma with curettage and bone grafting in our case series yielded a good result in general. Patient's symptoms were eliminated, the bone grafts incorporated well, and no recurrence occurred. However, there are some notable differences between the results of artificial bone substitutes and iliac crest autograft.

The use of artificial bone substitutes decreased the operating time and the duration of hospitalization when compared with harvesting iliac crest bone graft. The average operating time was cut down to 52 minutes from 76 minutes. The main reason was obviously because there was no need to harvest the iliac crest bone graft. There was also decreased blood loss. The risk of intraoperative contamination of the iliac crest wound was eliminated. Although our department performed all of the operations under general anaesthesia, it could theoretically be done under digital block with local anaesthesia if harvesting iliac crest bone graft was not needed. The risks of general anaesthesia could then be eliminated.

The average length of hospital stay of patients receiving artificial bone graft was 1.3 days, which was shorter as compared with an average of 2.75 days in patients receiving autograft. Within the autograft group, the average length of hospital stay was slightly shorter if the bone graft was harvested using a trephine biopsy needle (2 days) rather than the open method (3 days). Patients experienced postoperative iliac crest wound pain which delayed their ambulation. Redivac drain was inserted into the iliac crest wound in cases requiring open harvest of bone graft. Time needed to return to ambulation and drainage of the iliac crest hematoma contributed to the slightly prolonged hospital stay compared with patients receiving artificial bone substitutes. If trephine biopsy needle was used to obtain bone graft, the wound pain would be

reduced and a surgical drain was not necessary. However, it might be difficult to obtain enough bone graft using a biopsy needle when faced with a relatively sizeable lesion.

In our case series, autografts took an average of 5 weeks to consolidate. This result is comparable to that quoted in the literature. Huseyin et al⁸ reported that consolidation of iliac crest autograft took a mean of 38 days. Artificial bone substitutes, however, take a longer and more variable time to incorporate. Our patients require an average of 19 weeks for artificial bone graft incorporation. Jeffrey et al⁴ reported that demineralized bone matrix took 5 months for incorporation. Choy et al⁶ reported the mean time for calcium sulfate pellet remodelling was 10 weeks. However, the longer duration necessary for incorporation of artificial bone substitutes did not affect the final functional outcome of our patient or the healing rate of the bone graft. It implies that patient receiving artificial bone substitutes may need a longer-term radiographic follow-up.

Autograft is the standard treatment to fill up the bone void after curettage of enchondroma. It provides good osteogenicity, osteoinduction, and osteoconduction. The bone graft incorporation is fast and reliable. However, autograft supply is limited. It may also come with donor site morbidities, including wound pain, bleeding, hematoma, infection, fracture, sensory loss, abdominal herniation, and unsightly scar. Cosmetic problems over the iliac crest region especially concern the female patients.

Huseyin et al⁸ investigated the application of allograft in treating hand enchondromas, and evaluated the long-term results of both autograft and allograft application. They concluded that both groups yielded good functional and radiographic results. Although the allograft group took a longer time for consolidation (51 days) compared with the autograft group (38 days), the clinical effect is minimal. This result is expected as the morselized allografts only have osteoconductive effect but do not have osteogenic or osteoinductive properties. However, autografts contain all three properties. Allografts also come with other disadvantages, including immunological rejection and transmitting infective diseases.

DBX[®] Demineralized Bone Matrix is one of the various kinds of artificial bone substitutes used in our case series as well as reported in the literature. It is an osteoinductive and osteoconductive material composed of demineralized bone matrix from human donors mixed with collagen carrier. The demineralized bone matrix is produced by removing minerals from the cortical bone. It maintains the trabecular scaffold of the original allograft and contains osteoinductive growth factors. The bone morphogenic proteins (BMPs) are the active components in demineralized bone matrix which stimulate bone growth and regeneration. Demineralized bone matrix can also combine with other materials e.g., cancellous allograft, calcium sulfate to produce different form of composite products. Jeffrey et al⁴ reported the use of DBX[®] to fill up the bone void after curettage of an enchondroma. Radiographs showed good incorporation at 5 months after operation. In our series, the three patients who received DBX[®] had radiographic incorporation at an average of 21 weeks (range, 12–33 weeks). These results are comparable to the 6-month remodelling time as suggested by the manufacturer manual by Synthes.

Calcium phosphate bone cement is another artificial bone substitute used to treat enchondroma after curettage. Because it is in the form of a paste, it can fill up a bone void without any dead space. However, care must be taken not to let the bone cement leak out of the cavity, which may irritate the soft tissue and cause complications. A commercial form is Norian SRS (Synthes). It consists mainly of monocalcium phosphate and has high initial strength for early mobilization. It was used in one of our patients. Yasuda et al⁵ reported the usage of Biopex (Mitsubishi Materials

Corp, Tokyo, Japan), which was another product of bone cement to treat enchondroma after curettage. Biopex mainly consists of tri-calcium phosphate. It is prepared by mixing sterile white powder with a sterile solution until a doughy form was obtained. It was then injected as a paste form into the tumour cavity. Incorporation occurred at an average of 4.5 months after surgery, whereas the incorporation time was 12 weeks for our patient.

Choy et al⁶ used calcium sulfate pellet (Osteoset, Wright Medical Co., Memphis, TN, USA) in the treatment of enchondroma after curettage. Calcium sulfate serves as a scaffold for osteogenic cells and neovascularization for generating new bone. The grafted calcium sulfate acts as a space filler preventing invasion of other soft tissues. It is then gradually resorbed by subsequent bone remodelling. In their study, the calcium sulfate pellets were completely resorbed by postoperative 6 weeks, followed by bone remodelling at an average of postoperative 10 weeks. All patients achieved full range of motion except one with pathological fracture, and no recurrence or other complications were encountered.

Other substances have been used to fill the bone defects after curettage, including Plaster of Paris and hydroxyapatite. The use of Plaster of Paris was first described by Dreesman⁹ in 1892. The plaster was reabsorbed after a few weeks and replaced by bone. However, it came with the risk of reactions occurring during the contact with soft tissue and development of fistulas.¹⁰ It is no longer a popular choice. Hydroxyapatite application has also been described in the literature.¹¹ However, some patients complained of residual local pain, and this was attributed to the irritation by the material at the soft tissue.

In our hospital, enchondromas presenting with pathological fracture are all allowed to heal with splintage before definitive curettage and bone grafting are performed. This is the common school of thought in the literature, as concurrent intervention to the fracture and the enchondroma will increase the complication rate.⁸ However, this has the disadvantage of delaying the histological diagnosis and prolonging the disease process.¹ With the development of injectable calcium sulfate with high initial strength, Lin et al¹² reported the result of one-stage surgical treatment for enchondromas presenting with pathological fracture in 2013. They treated eight solitary enchondroma of a digit with undisplaced or minimally displaced pathological fracture. All fractures healed after a mean of 8 weeks after operation. The calcium sulfate cement was completely absorbed and replaced by newly formed bone at 12 weeks after operation. Although further studies are needed to determine whether injectable calcium sulfate can be applied in treating displaced and unstable pathological fractures, it shows that the development of newer and stronger artificial bone substitutes could potentially shift our treatment paradigm.

In summary, the application of artificial bone substitutes in the treatment of enchondroma is a sound alternative to iliac crest

autograft. Artificial bone substitutes yield good functional and radiographic outcome. Moreover, they can eliminate donor site morbidities, decrease operation time, and hospital stay. They are readily available and easy to use. Although artificial bone substitutes take a longer time to incorporate than iliac crest autograft radiographically, it does not affect the functional outcome or union rate.

This study is a small case series that concluded artificial bone substitutes are good alternatives to iliac crest autograft in treating hand enchondroma. Although we did not directly compare the results between different artificial bone substitutes, we found that DBX[®] putty was technically easy to apply and had good osteoinductive properties. A larger patient group and longer term follow-up for the efficacy and comparison between different artificial bone substitutes is needed.

Conflicts of interest

All authors have no conflicts of interest to declare.

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References

1. Markus F, Martin L. Retrospective review of outcome after surgical treatment of enchondromas in the hand. *Arch Orthop Trauma Surg* 2009;**129**:729–34.
2. Schwartz HS, Zimmerman NB, Simon MA, et al. The malignant potential of enchondromatosis. *J Bone Joint Surg Am* 1987;**69**(2):269–74.
3. Wulle C. On the treatment of enchondroma. *J Hand Surg (Br)* 1990;**15**:320–30.
4. Jeffrey VM, Osak OO, Jeffrey W. Treatment of benign bone tumours of the hand using demineralized bone matrix. *Plast Reconstr Surg* 2009;**123**(6):214e–6e.
5. Yasuda M, Masada K, Takeuchi E. Treatment of enchondroma of the hand with injectable calcium phosphate bone cement. *J Hand Surg Am* 2006;**31**:98–102.
6. Choy WS, Kim KJ, Lee SK, et al. Treatment for hand enchondroma with curettage and calcium sulfate pellet (OsteoSet) grafting. *Eur J Orthop Surg Traumatol* 2012;**22**:295–9.
7. Chinese QuickDASH questionnaire (QMH, Hong Kong version) translated by Chan KYR in 2006 from the DASH Outcome Measure (jointly developed by the Institute for Work & Health and the American Academy of Orthopaedic Surgeons).
8. Huseyin Y, Tackin O, Erhan C, et al. Long-term results of autograft and allograft applications in hand enchondromas. *Acta Orthop Traumatol Turc* 2004;**38**:337–42.
9. Dreesman H. Über Knochenplombierung. *Beitr Klin Chir* 1892;**9**:804–10 [in German].
10. Tay BK, Patel VV, Bradford DS. Calcium sulfate- and calcium phosphate-based bone substitutes. Mimicry of the mineral phase of bone. *Orthop Clin North Am* 1999;**30**:615–23.
11. Yamamoto T, Onga T, Marui T, et al. Use of hydroxyapatite to fill cavities after excision of benign bone tumours. Clinical results. *J Bone Joint Surg (Br)* 2000;**82**:1117–20.
12. Lin SY, Huang PJ, Huang HT, et al. An alternative technique for the management of phalangeal enchondromas with pathologic fractures. *J Hand Surg* 2013;**38A**:104–9.