INTRODUCTION

Osteoid osteoma (OO) is a benign osteoblastic bone tumor typically occurring in younger patients in their second and third decade of life. It predominantly affects males, though patients of all ages and genders can be affected.1 OO accounts for 10–12% of benign bone tumors.2–4 Approximately 50% occur in the lower extremity, namely, in the femur and tibia, whereas 10% occur in the hand and wrist.5

Clinically, OO causes focal tenderness, fusiform swelling if present in a finger, and a clubbing deformity of the nail when present in a distal phalanx.6,7 Nocturnal pain is a classic feature that responds to nonsteroidal antiinflammatory drugs.1,8,9 This is thought to be due to suppression of inflammatory mediators such as prostaglandins, which are highly concentrated in the OO nidus.10,11

The radiographic hallmark of OO is a nidus with lytic features and a sclerotic rim.12 However, OO is often difficult to diagnose using conventional radiography and can mimic other disease states such as chronic osteomyelitis.7 Bone scan (i.e., 99mTc scintigraphy), computed tomography imaging, or magnetic resonance imaging are often required to clarify the diagnosis.1 Despite these technological advancements in diagnostic imaging since the first report of OO by Jaffe13 in 1935, definitive diagnosis remains challenging.

CASE REPORT

A 41-year-old right-hand-dominant business professional presented in April 2015 with a 1-year history of painful swelling of the right long finger proximal phalanx. She recalls no history of any penetrating trauma. She had swelling centered at the associated proximal phalanx and proximal interphalangeal joint (PIPJ), which was progressively increasing for the last year. Pain had been partially controlled by NSAIDs. An ultrasound showed moderate synovitis and small effusion around the right long finger PIPJ. An x-ray demonstrated sclerosis and a hollowed-out lesion at the distal aspect of the proximal phalanx. Subsequent CT imaging showed a small button sequestrum measuring 2.4 × 2.4 mm in diameter on the volar aspect of the distal aspect of the proximal phalanx. (Fig. 1).
There was a surrounding radiolucency (of 1.6 mm) and loss of the anterior bony cortex with a diameter of 4.9 mm in keeping with a cavity. The radiologist’s report diagnosed these features as suspicious for chronic osteomyelitis.

Given the potential diagnosis of osteomyelitis and the patient’s failure to respond to conservative management (i.e., NSAIDs), she underwent exploration and debridement of the lesion in May 2015. A zigzag Brunner incision was made on the volar aspect of the long finger from the proximal base of the proximal phalanx to the distal interphalangeal joint. Skin flaps were raised and neurovascular bundles retracted. An area of soft-tissue swelling was noted on the proximal phalanx near the PIPJ, and thickened fluid was encountered. The flexor sheath was subsequently opened and serosanguinous fluid was expressed. Samples of this fluid were collected and sent for microbiology assessment. Further dissection past the fibrotic tissue of the flexor sheath revealed a round lesion on the neck of proximal phalanx. The lesion was cavitated with granulation tissue within the nidus (Fig. 2). This tissue was debrided with a curette and collected for pathology and microbiology. After debridement, the proximal phalanx had a shallow, circular defect approximately half the width of the bone. The wound was irrigated thoroughly with saline and a silastic drain was placed in the bony defect. The incision was closed in 1 layer using nylon suture. Saline compress and dry gauze were used for dressing. The hand was splinted in a functional position.

After surgery, fluid samples taken intraoperatively revealed no growth of microorganisms, fungus, mycobacteria, or acid fast bacilli. Microscopic examination of the tissue sample/sequestrum showed reactive osteoid with calcification. No sequestrum or dead bone was identified. Due to the contradiction of this finding with the CT report, a consultation was made to a pathologist specializing in musculoskeletal conditions who confirmed the diagnosis of OO. The specimen underwent hematoxylin and eosin staining. The pathologist report described a fragmented, sharply delineated central nidus that was composed of calcified osteoid. The nidus was lined by plump osteoblasts and growing within highly vascularized connective tissue with no signs of inflammation and surrounded by thick fragments of dense bone (Fig. 3).

The patient was seen in clinic 3 months postoperatively. The patient appeared to have an uneventful recovery with return of full function of the hand. However, 6 months postoperatively she started experiencing throbbing pain and swelling of the same digit. On examination, the pre-
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She underwent reexploration and excision of the lesion in January 2016. This was done in the same manner as the previous operation with thorough curettage of the lesion. Pathologic examination of the tissue confirmed the diagnosis of OO once again. She was seen in follow up at 1 month and 1 year postoperatively. In her last clinic visit in January 2017, she had full function of her hand. Other than occasional discomfort and scarring, no recurrence was detected clinically or radiographically.

**LITERATURE REVIEW**

**Methods**

We performed a comprehensive literature review on OO involving the bones of the hand and carpus. The purpose was to determine if there is a pattern in the distribution of OO throughout the bones of the hand and wrist. Based on the case series and case reports compiled from this search, we also aimed to summarize and highlight the epidemiology, natural history, clinical findings, diagnosis, and prognosis of this disease.

A title and abstract search was conducted using Medline and PubMed databases between June 1985 and January 5, 2017, for articles published on OO involving the bones of the hand. The following search terms were utilized: osteoid, osteoma, hand, finger, phalanx/phalanges, metacarpal, and carpal. Medical Subject Heading terms were employed where possible. The following inclusion criteria were used: (1) biopsy-proven OO of either a carpal, metacarpal, or phalangeal bone of the hand and (2) specification of a single, discrete bone involved in a case report or case series (e.g., middle phalanx of the third finger). Each biopsy-proven nidus was counted as 1 case of OO, regardless if more than 1 occurred in the same person, or even the same bone; however, recurrences in the same bone were not included. All studies included in the Results section that were not specifically referenced in the text of this study are included in Appendix 1.

![Fig. 4. X-ray (anteroposterior view) of the recurrent OO in the head of the right long finger proximal phalanx.](image)

![Fig. 5. A, MRI (anteroposterior view) of recurrent OO in right long finger proximal phalanx; B, MRI (sagittal view) of the same digit.](image)
RESULTS

Ninety studies published since 1985 yielded 289 cases of OO involving the bones of the hand and wrist (Table 1; Fig. 6). OO most commonly affected the phalanges (59.2%), followed by the carpal bones (30.1%), and then the metacarpals (10.7%; Table 2). OO most commonly affected the long finger ray with 34.7% of the 202 cases involving the rays of the hand (Table 3). The long finger ray also had the most cases of OO among all metacarpals, proximal phalanges, and distal phalanges. The least affected was the small finger ray (10.9%). Fifty-four percent of OO affecting the carpal bones occurred in the scaphoid and capitate (Fig. 6). The trapezium was the least commonly affected carpal bone with only 1 case of OO.

DISCUSSION

In this article, we reviewed an OO case that was initially misdiagnosed as osteomyelitis, one of several bony lesions known to mimic OO including Brodie’s abscess (i.e., intraosseous abscess), enchondroma, osteoblastoma, osteochondroma, osteosarcoma, and subungual exostosis.7,11 Misdiagnosis can delay proper management and unnecessarily prolong symptoms. Our case report highlights the risk of recurrence in this condition. This article also includes a literature review on the anatomic distribution of OO in the hand and wrist, which is the most comprehensive to date to our authors’ knowledge. A weakness of this review is that it was not systematic in nature, and therefore some case reports may not have been included in our analysis of 90 studies. A thorough review of the literature reveals that there is still no clear mechanism for the development of OO in the hand. The implication of trauma in OO development has been a controversial point for decades.19 A history of trauma was reported in 18–50% of hand and wrist OO cases,1,4,11,20,21 although the diagnosis may sometimes be confused for stress fracture or capsular strain.22 Abnormal blood markers were not noted in any OO cases where blood was analyzed.1,4,11,20

According to a retrospective review of 37 OO cases by Simon et al.,23 OO accounts for 5.9% of all hand tumors and the most common locations involved the phalanges (59.5%), metacarpals (24.3%), and lastly the carpals (16.2%). Our literature review results found agreement with Simon et al.23 with respect to phalangeal involvement being most common at 59.2% (Table 2). However, based on our review of 289 cases, we found that carpals were involved more frequently than metacarpals at 30.1% and 10.7%, respectively.

OO cases tended to occur more frequently near the midline axis of the hand, a longitudinal line through the long finger ray, capitate, and scaphoid (Fig. 6; Table 3). This line is also known as the reference line for abduction and adduction of the hand.24 The incidence of OO in the hand appears to decrease with increasing distance from the midline axis of the hand in the coronal plane. There is no explanation in the literature for why this may be. This finding warrants future investigation as to a potential mechanism for the distribution of OO in the hand.

OO in the hand and wrist is most common in young adults, and the average age of affected individuals in several larger studies was 23–35 years.1,4,11,20,21,25–27 However, any age is possible ranging from reports of congenital OO28 to 1 case of a 70-year-old male being affected.29 Although OO in general is frequently cited as being 2 to 3 times more common

| Table 1. Number of OO Cases per Study Included in Literature Review (90 Studies, 289 Total OO Cases) |
| Study | Number of OO Cases | Study | Number of OO Cases | Study | Number of OO Cases |
|-------|--------------------|-------|--------------------|-------|--------------------|
| Claeys et al., 2016 | 1 | Amrami and Berger, 2006 | 1 | Bednar et al., 1993 | 19 |
| Durgia et al., 2016 | 1 | Laffosse et al., 2006 | 2 | Glickman et al., 1993 | 1 |
| Park et al., 2016 | 1 | Giraud et al., 2005 | 1 | Lamme et al., 1993 | 1 |
| Cakar et al., 2015 | 1 | Ramos et al., 14 | 1 | Zara et al., 1993 | 1 |
| Güner et al., 2015 | 1 | Themistocleous et al., 2005 | 1 | Brown et al., 1992 | 1 |
| Gupta et al., 2015 | 1 | Bilgin et al., 2010 | 8 | Chamberlain et al., 1992 | 1 |
| Hamdi et al. | 17 | Burger and McCarthy | 7 | Oosterbosch et al., 2011 | 1 |
| Kotnis and James | 7 | Ilaian et al., 2004 | 1 | Muren et al., 2012 | 3 |
| Kussman et al., 2015 | 1 | Ramesh et al., 2004 | 2 | Helzel and Kreisköther, 1990 | 1 |
| Salva-Coll and Terrades-Cladera, 2015 | 1 | Arora et al., 2018 | 1 | Tricoire et al., 1991 | 1 |
| Papachristos and Pasparakis, 2014 | 1 | Marcuze et al., 2014 | 9 | Zanasi et al., 1990 | 1 |
| Taylor et al., 2014 | 1 | Niame et al., 2002 | 2 | Chen and Caplan, 1989 | 1 |
| Jafar et al. | 24 | Olmedo-Garcia et al., 2002 | 1 | Meng and Watt, 1989 | 2 |
| Aghotane and El Fezzazi, 2012 | 1 | Schindler et al., 2002 | 1 | Walker and Meats, 1989 | 1 |
| Becc et al. | 1 | Uda et al., 2012 | 1 | Allieu et al., 1988 | 46 |
| Ozbek et al., 2011 | 1 | Araz et al., 2001 | 2 | Crosby and Murphy, 1988 | 1 |
| Ek and McCullough | 12 | De Smet, 1987 | 15 | Kozlowski et al., 1988 | 6 |
| Akhlaghoor et al. | 2 | Basu et al., 1999 | 1 | Mark et al., 1988 | 3 |
| Harrod et al., 2010 | 1 | Inagaki and Inoue, 1999 | 1 | Nicolaisen et al., 1988 | 1 |
| Herzberg et al., 2010 | 1 | Kreiter et al., 1999 | 1 | Foucher et al., 1987 | 4 |
| Tsang and Wu | 26 | Mayer et al., 1999 | 1 | McCartney et al., 1991 | 1 |
| Jackson and Markiewitw, 2008 | 1 | Rozier et al., 1998 | 1 | Ambrosia et al., 2011 | 19 |
| Derks et al., 2008 | 1 | Hartmann et al., 1997 | 1 | Bowen et al., 2006 | 2 |
| Di Gennaro et al. | 1 | Rex et al., 1997 | 1 | Chevrot et al., 1997 | 1 |
| Hedrich et al., 2008 | 2 | Soler et al., 1997 | 2 | Shaw, 1987 | 1 |
| Malik et al., 2008 | 1 | Zanetti et al., 1997 | 1 | Mullin et al., 1986 | 1 |
| Zouari et al., 2008 | 8 | Lisan et al., 1996 | 3 | Nunez-Samper et al., 1986 | 1 |
| Eroszlu, 2007 | 2 | Georgoulis et al., 1995 | 3 | Doyle et al., 2011 | 5 |
| Chronopoulos et al. | 10 | Nakanishi et al., 1994 | 1 | Szabo and Smith, 1989 | 1 |
| Messoudi et al. | 27 | Wacht et al., 1995 | 1 | Kernohan et al., 1985 | 2 |

References provided in “name and date” format in this table are listed in Appendix 1.
in males,\textsuperscript{1,25} the average of 10 large case series over the last 30 years shows a more equivalent sex ratio of 1.3:1 (male:female) for OO cases in the hand and wrist.\textsuperscript{1,3,4,11,20,21,25–27,29} Based on the data available, the right hand appears to be involved 1.9 times more frequently than the left.\textsuperscript{1,3,4,20,21}

The diagnosis of OO in the hands can be challenging due to clinical findings overlapping with synovitis and arthritis.\textsuperscript{4} The average time to diagnosis is approximately 13–20 months.\textsuperscript{4,20,21,26} Two particularly distinctive features that may lead to earlier diagnosis are nocturnal pain and pain relieved by NSAIDs, particularly salicylates. Nocturnal pain is present in 50–84\% of cases,\textsuperscript{1,21} whereas pain responding to NSAIDs was reported in 70\% of cases on average.\textsuperscript{1,11,20,21}

Only 52\% of hand and wrist cases of OO are diagnosed on radiographic analysis on average, due to their nonspecific appearance in this modality.\textsuperscript{1,4,11,20,25,27} A more reliable modality is \textsuperscript{99m}Tc scintigraphy, which can diagnose OO in 91\% of cases on average.\textsuperscript{1,4,11,21} but may lack specificity due to diffuse isotope uptake in surrounding tissues.\textsuperscript{1} MRI and CT imaging are successful in recognizing OO in all cases, though CT imaging was used 41\% more often.\textsuperscript{1,4,11,27} CT is considered the most specific imaging modality for identifying OO, given the sensitivity of MRI for registering soft-tissue reaction surrounding the nidus that may confuse the diagnosis.\textsuperscript{30} Of course, histology is the definitive diagnosis for all cases of OO in the hands and wrist.\textsuperscript{1,4,20,51}

Our case report illustrates the difficulty of differentiating between osteomyelitis and OO both clinically and radiologically. A consultation with a specialist musculoskeletal pathologist was required to confirm the diagnosis.

Management of OO of the hand and wrist showed great diversity across studies in this literature review, varying from en bloc resection, excisional biopsy, and curettage with or without bone grafting.\textsuperscript{1,4,20,27} Excision by curettage is thought to be incomplete in comparison with en bloc resection and may increase the chance of recurrence.\textsuperscript{32,53} The recurrence rate for OO treated with surgery was approximately 12\% in the larger case series where it was reported.\textsuperscript{1,11,20,26} Given the heterogeneity in reporting of surgical technique among the studies analyzed, recurrence rates between techniques could not be compared reliably. Incomplete excision may be associated with a greater need for bone grafting due to the greater likelihood of repeat excision with wider margins, which may increase the risk of postoperative fracture without bone grafting.\textsuperscript{21}

### CONCLUSIONS

There is no consensus in the literature to explain the pathophysiology for OO development in the hand. Our literature review showed a predilection for involvement of the third ray, as well as the scaphoid and capitate bone: all continuous with the midline axis of the hand. Future studies will be required to elucidate the mechanism behind OO formation. En bloc resection and excision by curettage were the most common surgical treatments, with the former being considered to have a lower risk for disease recurrence. In general, OO of the hand and wrist should be suspected in adults younger than 40 years with chronic, focal pain that is worse at night, relieved by NSAIDs, and is otherwise unexplained.
ACKNOWLEDGMENTS
We would like to thank Jessica Murphy for her administrative assistance in this project.

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