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A Bizarre Parosteal Osteochondromatous Proliferation – Case Report and Review of Similar Hand Lesions Recorded Since 1983

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SUMMARY

Bizarre parosteal osteochondromatous proliferation (BPOP) is an unusual lesion mostly affecting the bones of the hand. The mass grows from the bone surface and consists of cartilaginous, osseous and fibrous tissue. The lesion is commonly under/misdiagnosed and confused with other lesions, mostly the osteochondromas. We present a patient with BPOP that initially confused the practitioner and radiologist in their diagnosis. We discuss the clinical, radiologic and histologic characteristics of BPOP of the hand since its first report in 1983 and present its main differential diagnosis. We reviewed 184 cases. Female were affected in 52% and male in 48%. Proximal phalanges were most commonly affected, followed by middle phalanges and metacarpals. The pain was reported in 47,9 % of all reported papers. The most common surgical treatment was surgical excision, and the rate of recurrence was 47.3%.

Key words: benign lesion, bone, cartilage, hand

BACKGROUND

Bizarre parosteal osteochondromatous proliferation (BPOP) is a tumor-like lesion described by Nora et al. in 1983 [1]. Nora´s lesion, as it is sometimes called, grows most commonly from the small bones of the hand [1,2]. The lesion is usually clinically characterized as a painless lump growing to a maximum of 3.0 cm in diameter [2]. Some authors suggest that BPOP arises from injured periosteum and that it is a reparative process, while recent studies are suggesting a neoplastic character due to the presence of chromosomal changes [3,4].

To our knowledge, only 48 published papers with 184 cases of BPOP involving the hand (metacarpals and phalanx) have been reported in the literature (Table 1). In our clinical report, a case of Nora's lesion is presented in a patient with osteoarthritis of the distal interphalangeal joints which led to initial clinical and radiological misdiagnosis of the lesion due to its rarity. The lesion was eventually histologically confirmed. In this article, we review and discuss epidemiological, clinical, radiographic and histological data of all BPOP lesions of the hand since its first description in 1983 and discuss its main differential diagnosis.

CASE REPORT

A 54-year-old male presented to the rheumatology outpatient clinic with a six-month history of a fast-growing tumorous mass over the middle part of the right index finger. He was referred by a general practitioner, who suggested a peculiar progression of the nodal arthritis lesion. The patient presented only a little discomfort without pain, previous trauma or surgery.

On physical examination, Heberden's nodes were found and the mass over the index finger, which was stiff to palpation and measured 20 x 10 x 5 mm. The range of motion of PIP and DIP joints was not impaired. Neurovascular functions were normal.

A plain radiograph of a right hand demonstrated third-degree osteoarthritis in the DIP joints and a dense mass extending from the radial aspect of the second phalanx (Fig. 1). There was no abnormality of the un-

Tab. 1. Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

Study	Gender	Age	Localization	Symptoms	History of trauma	Radiography	Histology	Treatment	Recurrence
Nora et al. (1983) [1]	16 F 19 M (lesions of the hand and feet together, no detailed info)	14-74 (median of 34) (hand and feet together)	31 in hand: 3 x proximal phalanx (not otherwise specified) 4x proximal phalanx of thumb 4x proximal phalanx of index finger 6x proximal phalanx of third finger 3x middle phalanx of index finger 4x middle phalanx of third finger 1x middle phalanx of fourth finger 2x second metacarpal 1x third metacarpal 3x fifth metacarpal	in 18 cases: (hand and feet together) 3 patients with no symptoms 5 patients with tenderness (2 with rapid growth) others with discomfort, tumor formation	none of the patients	-	BPOP	26 cases by excision some by amputation (not specified)	18 cases recurred once (8 of those recurred twice)
Davies (1985) [5]	F	65	proximal inter-phalangeal joint of the index finger	persistent pain, insufficient range of motion	yes	-	BPOP	excision	no
Lindeque et al. (1990) [6]	F	45	proximal phalanx of the left thumb	nodular swelling	no data	-	1st: low grade osteosarcoma 2nd:BPOP	1st: excision 2nd : wide excision 3rd: skin was removed en bloc with the tumor + cryotherapy	1st 2 months later 2nd recurrence

Tab. 1 (cont.). Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

Teoh et al. (1992) [7]	M	38	middle phalanx of the middle finger	progressive enlarging lump, tender, no pain	yes	-	BPOP with loss of chondro - cyte column formation and loss of lacunar architecture of the chondrocytes	excision	4 months after surgery
Wu et al. (1992) [8]	F	27	proximal phalanx of the middle finger	painful swelling	no	-	BPOP	1.excision 2.excision 3.excision	18 months after first excision
Meneses et al. (1993) [9]	F (34) M (31) involving also other sites	8-73 median 33.9	36 patients involving hands (exact location not known)	Swelling pain (3 patients)	yes (in 8 patients)	-	BPOP	excision (21 patients) wide excision (3) bone graft (2) amputation (2)	22 patients (no data informati on)
Derrick et al. (1994) [10]	M	37	distal phalanx of the thumb	tender, crusted area in the centre, swelling	no	-	BPOP	excision	24 months later
Breidahl et Wylie (1995) [11]	F	31	fifth metacarpal	non-tender mass	yes	-	first : osteo-chondroma second:BPOP	1st: excision 2nd: excision	3 months after first excision
Smith et al. (1996) [12]	7 patients (6 M, 1 F) 3 cases involving hand	18-37 (median 30)	1. little finger phalanx 2. no data 3. no data	4 patients reported pain	yes (5 cases) no (2 cases)	-	BPOP	all by local excision in 3 patients afer recurrence: wide excision	3 patients
Tannenba um et al. (1997) [13]	M	44	fifth digit proximal to the PIP joint	painless growth	no	-	BPOP	excision	no
Chuang et al. (1997) [14]	F	47	proximal phalanx of right index finger	pain and swelling	no	-	BPOP	intraoperative frozen section of bone tumor followed by resection	no
Campa-nacci et al. (1999) [15]	F	35	fifth metacarpal	local sweling with little or no pain	50% of patients reported trauma	-	BPOP	wide excision	no
	?	?	fifth metacarpal						
	?	?	proximal phalanx of the first finger						
	?	?	fifth finger						
Garcia-Alvarez et al. (1999) [16]	F	32	second phalanx of the fifth finger	painless	?	-	fibroplastic proliferation with trans-formation into chondro-osseous tissue	1st: extirpation 2nd: excision with underlying bone cortex	1 month later
Torre-ggiani et al. (2001) [17]	M	35	proximal phalanx of the thumb	painless lesion, restriction to motion	no	-	BPOP	removal	6 months later
De Smet et al. (2001) [18]	F	45	proximal phalanx of the left thumb	slightly tender	?	-	BPOP	excision	no
	M	18	distal phalanx of the left ring finger	skin was inflamed, reddish and warm	?	-		excision	no

Tab. 1 (cont.). Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

	M	34	proximal phalanx of the index finger	painless	no	-	BPOP	?	no
	F	62	fifth metacarpal	painless	no			?	no
	F	55	distal phalanx of the index finger	painful	no			?	no
	M	23	middle phalanx of the index finger	painless	no			?	no
	M	34	fifth metacarpal	painless	yes			?	no
	F	63	middle phalanx of the index finger	painless	no			?	6 months later
	M	43	proximal phalanx of the fifth finger	painless	no			?	no
	F	25	fifth metacarpal	painless	no			?	no
	F	33	middle phalanx of the small finger	painless	no			?	2 years later
	F	45	proximal phalanx of the middle finger	painful	no			?	3 weeks later
Orui et al. (2002) [19]	F	69	fifth metacarpal head	swelling	no	with intramedullary inflammatory extension by MRI	BPOP	excision	no
Godbee et Griffiths (2002) [20]	F	51	tip of the thumb	slowly enlarging mass, painless	yes	-	BPOP	excision	?
Soon et al. (2003) [21]	M	36	third metacarpo-phalangeal joint	hard mass, painless	no	-	1st:osteochondroma 2nd: BPOP	excision	5 months later
Claude et al. (2003) [22]	4 M (not specified)	?	middle phalanx of third finger (others not specified)	?	no	-	BPOP	excision	?
Agarwal et al. (2003) [23]	M	21	proximal phalanx of the little finger	swelling	no	-	BPOP	excision	no
Nilsson et al. (2004) [24]	M	39	? third finger	no data	no data	-	BPOP	excision	?
	F	24	? first finger						?
	F	46	? fifth finger						3 years later
Michelsen et al. (2004) [25]	M	34	proximal phalanx of index finger	painless, nontender mass	no	in 2 cases the margin of the lesion was not clearly defined	BPOP	excision with overlying pseudocapsule, if there was cortical erosion - the underlying cortex was excised	lost
	F	55	distal phalanx of the index finger	painful and tender	no				no
	F	62	fifth metacarpal	painless nontender mass	no				no
	M	23	middle phalanx of the index finger	painless nontender mass	no				no
	M	34	fifth metacarpal	painless nontender mass	yes				no
	F	63	middle phalanx of the index finger	painless nontender mass	no				no
	M	43	proximal phalanx of the little finger	painless nontender mass	no				no

Tab. 1 (cont.). Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

Michelsen et al. (2004) [25]	F	25	fifth metacarpal	painless nontender mass	no				no
	F	33	middle phalanx of the little finger	painless nontender mass	no				yes
	F	45	proximal phalanx of the middle finger	painful and tender	no				no
Rampoldi et al. (2005) [26]	F	46	fourth metacarpal bone	swelling, non-tender mass, little pain	no	-	BPOP	excision	no
	M	42	second phalanx of the fifth finger	swelling with mild pain		-	BPOP	excision	no
Le Bellec et Asfazadourian (2005) [27]	M	35	proximal inter-phalangeal joint of the middle finger	hard swelling	no	-	BPOP	1st:excision 2nd: excision	1st: 2 years later 2nd: no
	M	60	proximal phalanx of the fourth finger	painless swelling, painful later	no	-	BPOP	excision	no
Dhondt et al. (2006) [28]	M (15) F (9)	Mean age 38.8 (12- 81)	6 x proximal phalanx ? 11 x midle phalanx ? 5 x distal phalanx ? 2 metacarpals ?	?	?	-		resection	7 cases of recurrence 6 months after resection
Kraft et Hailer (2006) [29]	F	12	second metacarpal	painless swelling	no	-	BPOP	1st: marginal resection 2nd: autologous fibula grafting	2 years later
Lam et al. (2006) [30]	M	?	middle phalanx of the middle finger	painful, mild tenderness	no	-	BPOP	?	?
Makhson et al. (2008) [31]	M	76	proximal inter-phalangeal joint of the fifth finger	painless tumor	yes	appeared as chondrosarcoma	BPOP	wedge resection	?
Moretti et al. (2008) [32]	M	44	distal phalanx of the fifth finger	painful swelling (1992), relapse (2002), relapse (2004)	?	-	chondroma (2002) BPOP (2004)	amputation	?
Gruber et al. (2008) [33]	F	51	proximal phalanx of the thumb	painful mass	no	-	BPOP	intralesional excision	4 months later
	M	37	proximal phalanx of the third finger	discomfort	no			intralesional excision	no
Gursel et al. (2008) [34]	M	32	middle phalanx of the little finger	pain, limitation of range of motion	yes	-	BPOP	amputation of left fifth ray	no
Shapeero et Odone (2010) [35]	M	24	middle phalanx of the fifth finger	mildly painful, hard lump	yes	-	BPOP	resection	?
Chamberlain et al. (2010) [36]	F	39	third metacarpal	pain	?	-	1st read as an osteochondroma (different institution), 2nd read as BPOP (author's institution)	1st:resection (different surgeon) 2nd:wide excision + grafting (author)	1st :5 months later no after wide excision

Tab. 1 (cont.). Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

Chamberlain et al. (2010) [36]	M	38	middle phalanx of the middle finger	swelling, minimally painful	?	-	1st read: as an osteochondroma (different institution) 2nd read: as BPOP	1st:excision (different surgeon) 2nd:excision (author)	17 months after first resection
Jafari et al. (2010) [37]	F	31	proximal and middle phalanx of the ring finger	painful mass, pink skin, tender	yes	-	BPOP	no excision, only biopsy; spontaneous resolution	no recurrence at 20 months after onset
Berber et al. (2011) [38]	M 14 F 8 9 patients involved metacarpals (no phalax)	6-66 mean age 31,8	9 metacarpals 1. fourth metacarpal 2-9 (no data)	painless swelling (3) painful (7) limited motion (1)	yes (no specified which one)	1 case with medullary continuity in phalanx ????	BPOP	excision, in one metacarpal with reconstruction using a fibular strut graft	yes (6 patients) not specified
Cigna et al. (2011) [39]	F	47	distal phalanx of the thumb	progressive enlarging tumor	?	-	BPOP	resection	no
Joseph et al. (2011) [40]	F	29	middle phalanx of the little finger	localized swelling,	no	-	BPOP	marginal resection	18 months later
	M	41	middle phalanx of the ring finger	localized swelling	no	-		intralesional curettage	84 months later
	F	47	middle phalanx of the little finger	localized swelling	no	unable to exclude intramedullary involvement		marginal resection	no
	F	25	middle phalanx of the little finger	localized swelling, pain	no	-		marginal resection	25 months later
	F	65	distal phalanx of the middle finger	localized swelling	no	-		marginal resection	53 months later
	F	39	second metacarpal	localized swelling	no	-		marginal resection	no
	M	49	proximal phalanx of the index finger	localized swelling, pain	no	-		marginal resection	25 months later
	M	44	proximal phalanx of the ring finger	localized swelling, pain	no	-		marginal resection	no
	M	58	middle phalanx of the ring finger	localized swelling	no	-		marginal resection	no
Sakamoto et al. (2011) [41]	F	59	distal ring finger, ulnar side at the level of the distal phalangeal joint	swelling	no	continuity with the underlying bone marrow (CT)	BPOP	resection	no
Chaabane et al. (2011) [42]	M	52	middle phalanx of the index finger	hard mass	no	-	BPOP	excision	no
	F	24	middle phalanx of the middle finger	hard mass	no	-		excision	no
	F	45	distal phalanx of the middle finger	swelling and pain	?	-		excision	no
Barrera-Ochoa et al. (2012) [43]	F	39	third metacarpal	pain, increased swelling, and progressive limitation of range of motion	no	1. prior to initial resection - lesion with no medullary continuity (x-ray, MRI) 2. ossified mass, evident continuity between the medullary cavity (CT, MR)	BPOP	1st: resection 2nd:amputation of right third ray (author)	no

Tab. 1 (cont.). Reported cases of bizarre parosteal osteochondromatous proliferation of the hand from 1983 to 2016

Lynch et al. (2013) [44]	M	35	proximal phalanx of the right index finger	non-painful mass	no	-	no definitive cartilaginous cap	excision	no
Rana et al. (2013) [45]	F	45	proximal phalanx of the thumb	swelling	no	-	BPOP	excision	no
Kolbenschlag et al. (2013) [46]	F	29	distal phalanx of the thumb,	swelling, pain	no	-	BPOP	1st: excision (6 months earlier, different institution) 2nd: resection (author's institution)	no
Konijnendijk et Soepboer (2013) [47]	No data	22	middle phalanx of the second digit	swelling, mildly tender	no	-	?	?	?
Kumar et al. (2014) [48]	F	4	middle phalanx of the middle finger	pain and swelling	no	-	BPOP	excision	no
Rappaport et al. (2014) [49]	M	34	middle phalanx of the fourth finger,	hard lump	yes	-	BPOP	excision	?
Esenwein et al. (2015) [50]	F	43	middle phalanx of the little finger,	hard mass	no	-	BPOP	excision	no
total: **48 papers**	**total available : 100** **F=52 M=48**	**total available : 76 average= 40.3 (4-76) median= 39**	**184 cases** **102 well described**	pain reported in 23 papers (47.9 %)		cortico-medullary continuity reported in **3 cases** **1 representative**			**n=74** **yes=35 (47.3%)** **no=39 (52.7%)**

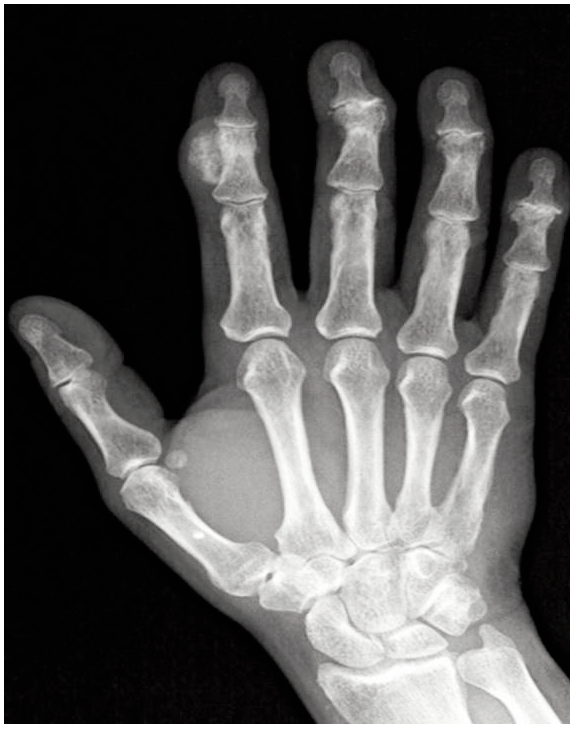


Fig. 1. Plain radiograph of the right hand showing a lobulated, calcified mass attached to the radial aspect of the second phalanx of the index finger; no bony architecture abnormality of the underlying phalanx; osteoarthritis of the DIP joints of the second to fifth fingers

derlying bony architecture, and cortical erosion was not detected. The radiologist suggested an osteochondromatous lesion.

The patient with these findings was presented to our outpatient clinic. Computed tomography (CT) and magnetic resonance imaging (MRI) were performed. CT confirmed an osseous, irregularly oval mass with sharp contours attached to the cortex of the medial phalanx extending dorsally and radially and the absence of medullary continuity between the lesion and the underlying bone (Fig. 2). The mass was nonhomogeneous on both T1- and PD-weighted images with

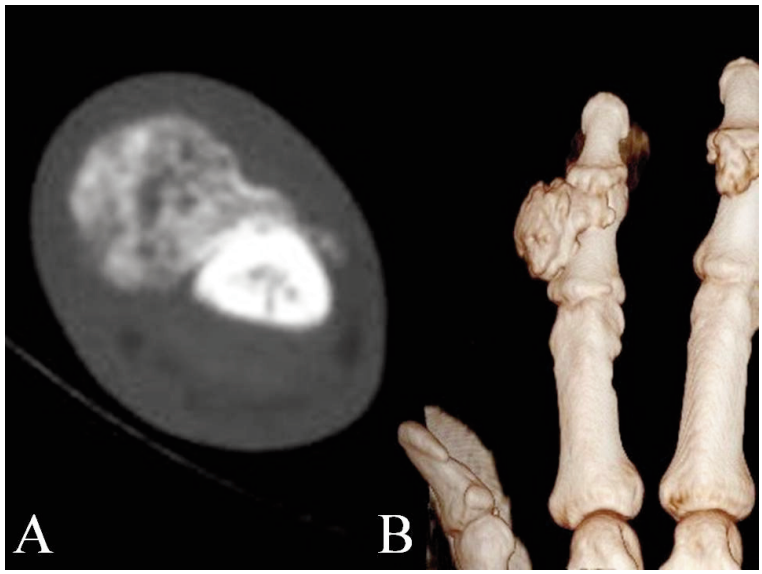


Fig. 2. CT scan of the right hand – (A) transverse, (B) MPR reconstruction: osseous, irregularly oval mass with sharp contours attached to the cortex of a medial phalanx extending dorsally and radially; no medullary continuity is seen between the lesion and the underlying bone; no signs of cortical involvement

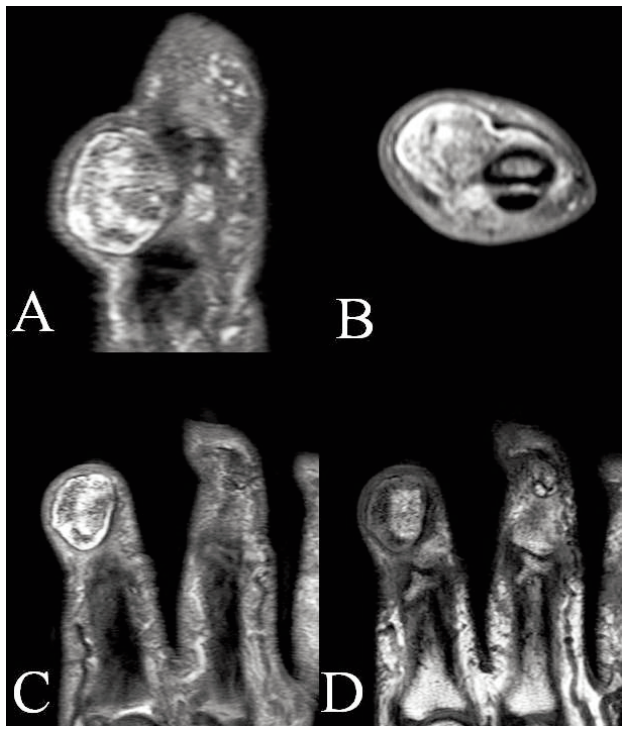


Fig. 3. Magnetic resonance imaging – (A) PDW sagittal, (B) PDW axial, (C) PDW HR coronal and (D) T1W coronal images – non-homogeneous oval lesion with thin non-signal surface layer attached to a medial phalanx of the second finger of the right hand; its peripheral and radial part is mostly hypointense on both T1- and PD-weighted images, its central and ulnar part is mostly hyperintense on both T1- and PD-weighted images; no signs of cortical or medullar involvement of the underlying phalanx

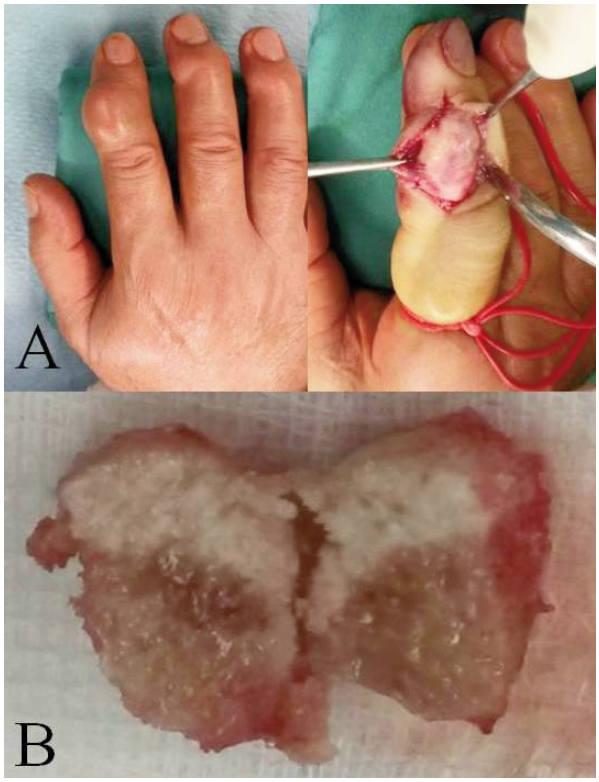


Fig..4. Pre-operative clinical presentation of the mass and dorsolateral incision (A) through which the mass was excised and the pseudocapsule was removed. Transection of the mass (B) showed two parts - one bony-like (down) and second cartilage-like (up) part

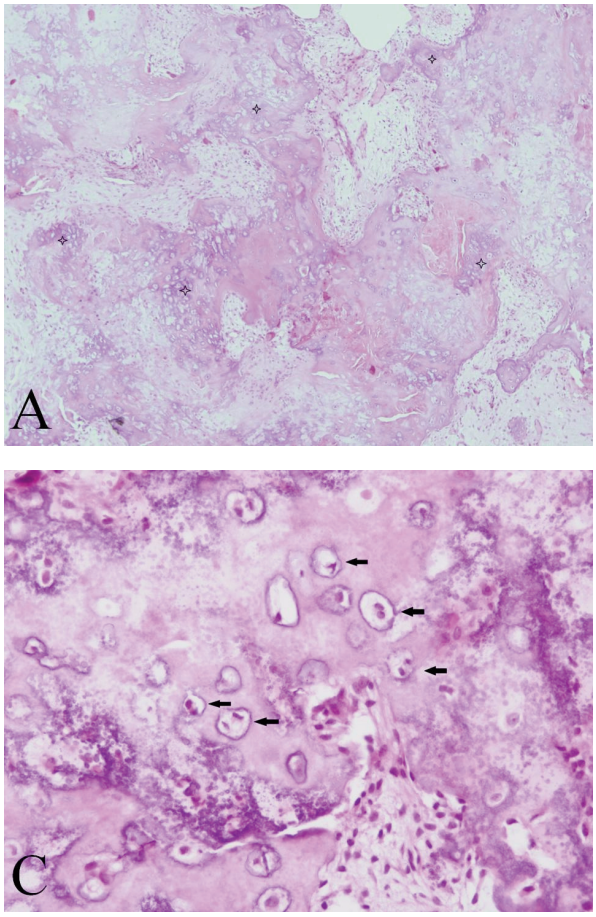
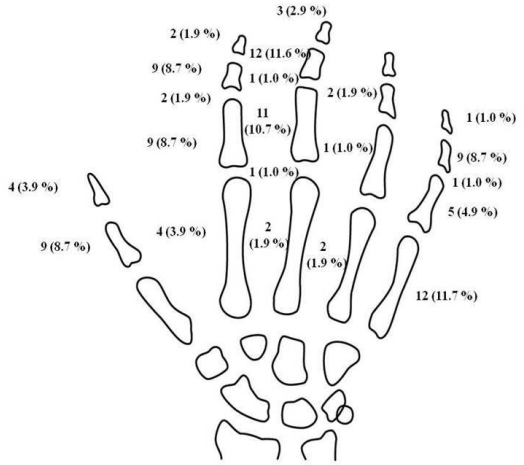


Fig. 5. Histology – A: (HE hematoxylin and eosin stain x40); Presence of disorganized endochondral ossification and irregular maturation of cartilage in bone produces chondro-osteoid with characteristic blue quality ("blue bone" – signs) and intertrabecular spindle cell proliferation. B: (HEx100); Chondro-osteoid with characteristic blue quality ("blue bone"), contains enlarged, bizarre, atypical chondrocytes (arrows), with maturation into bone. C: (HEx200); Enlarged, bizarre, hyperchromatic, atypical, often binucleated chondrocytes (arrows)



Site	n/%
DP	10/9.8
DIPJ	0
MP	32/31.4
PIPJ	4/3.9
PP	35/34.3
MPJ	1/1
M	20/19.6
total	**102**

Fig. 6. Distribution of all available BPOP lesions of the hand since 1983. M, metacarpals; MPJ, metatarsophalangeal joint; PP, proximal phalanx; PIPJ, proximal interphalangeal joint; MP, middle phalanx; DIPJ, distal interphalangeal joint; DP, distal phalanx

a thin no-signal surface layer. Its peripheral and radial part were mostly hypointense on both T1- and PD-weighted images, and its central and ulnar part was predominantly hyperintense on both T1- and PD-weighted images. There were no signs of cortical or medullary involvement (Fig. 3).

Based on the clinical and radiology results, a diagnosis of BPOP was considered and excision was indicated. The patient underwent surgery under local anesthesia. The mass was excised trough dorsolateral incision (Fig. 4a). An outgrowth from the cortical surface of the underlying bone of middle (second) finger was interrupted and excised. The pseudocapsule was removed. The architecture of the underlying bone was preserved without cortical violation. Transection of the mass showed two distinct parts: a brown, bone-like part and a white, cartilage-like part (Fig. 4b). Histological examination confirmed the initial suspicion of BPOP and excluded a malignancy (Fig. 5a, b, c). The patient was informed about a high rate of recurrence, but after 10 months there was no radiological and clinical recurrence.

DISCUSSION

BPOP is an uncommon lesion, and true prevalence is difficult to assess because most lesions are reported as case studies [2,9]. To our knowledge, exactly 184 cases of BPOP lesions involving hand have been presented in the literature since 1983, corresponding to a rate of 5.8 cases per year.

The mass may be confused with various benign and malignant conditions. The main differential diagnosis involves osteochondromas. Other entities that are similar in various sections include callus formation, florid reactive periostitis, turret exostosis, myositis ossificans and parosteal osteosarcoma. Analyzing all reported cases of BPOP of the hand, we review its key features and how to distinguish it from other lesions.

Clinical presentation

The mean age of the patients with a BPOP lesion of the hand is 40.3 years (52% females, 48% males, Tab. 1). In all reported cases of the hand (Tab. 1), it was possible to determine the specific location in 102 cases (Fig. 6). Proximal phalanges (n=35) are most commonly affected, followed by middle phalanges (n=32) and metacarpals (n=20). The BPOP mass is usually painless and grows over months to years [51]. This is similar to our case; however, analyzing all collected data, the pain was reported in 23 (48%) of the papers.

Osteochondromas are most commonly diagnosed in the second and third decades and more likely characteristic for metaphyses of long bones, although short bones are occasionally affected [52], similarly as in BPOP lesions which have recently been more frequently described adjacent to long bones [51,53]. Callus formation may occur at any age and is usually due to a stress fracture presenting with pain unless the fracture heals, which typically takes a few weeks. Florid reactive periostitis, just as BPOP, often affects small bones of the hands and occurs due to periosteal injury or without it [54]. Turret exostosis (ossifying hematoma) arises after an open wound through the extensor mechanism of the fingers causing a small pe-

riosteal defect with hematoma, which gradually ossifies and becomes mature, usually six months after the original injury [55].

Traumatic etiology in BPOP may or may not be present, as in florid reactive periostitis, and is not so clear as in turret exostosis. Myositis ossificans is a benign condition of heterotopic bone and usually occurs after muscle injury affecting subcutaneous and muscle tissue [56]. It is located in the larger muscles near long bones and is rarely located in the hand. Parosteal osteosarcoma is a very rare, painless, long growing malignant tumor usually affecting the metaphyseal region of long bones such as the femur, tibia, humerus and rarely found in the hands [57]. Therefore, it may be considered more important in the differential diagnosis of tibial and femoral BPOP lesions.

The above-mentioned conditions are characterised by similar patient age, gender, location and etiology and, therefore, this similarity is not pathognomic for diagnosis of BPOP.

Imaging

Radiographically, the lesions present as an osseous lump along the cortical surface of the bone similarly as in our patient. The underlying bone usually shows no evidence of medullary invasion, which is the main differential feature of an osteochondroma [3]. However, cases have been described where histologically proven BPOP lesions demonstrated communication of the lesion with the underlying medullary cavity on radiographs or CT [2,3]. We found that only three papers since 1983 described medullary continuity (Tab. 1). However, Berber et al. described medullary continuity in one case in a phalanx while there was no phalanx involved in their study and there were only nine metacarpals [38]. Barrera-Ochoa et al. found evident continuity between the medullary cavity of the third metacarpal [43]. However, before initial resection, there was no medullary continuity on MRI. Sakamoto et al. described one case of BPOP arising from the distal part of the middle phalanx with medullary continuity [41]. However, a clear zone between the lesion and the host bone was observed.

Radiographically, osteochondroma presents with a stalk of the underlying cortex and typically shows cortico-medullary continuity. If there is no evidence of medullary invasion, BPOP could be suggested [3]. Myositis ossificans is a soft tissue lesion usually not attached to the cortex. However, in half of the cases, the ossifications may be deeper and adhere to the periosteum and such lesions are known as parosteal myositis ossificans [58]. Radiographically, there is a characteristic "zoning" phenomenon characterized by a peripheral rim of calcification surrounding a central zone of lucency [59]. These radiographic findings are similar to the early stage of BPOP, and therefore it must be distinguished by histology. The finding of a fracture line with periosteal reaction confirms the diagnosis of callus formation and rules out BPOP, and other investigations are usually not required. Florid reactive periostitis appears as soft tissue calcification with periosteal reaction, which is absent in Nora´s lesion. Turret exostosis appears as a smooth, dome-shaped, extracortical collection of subperiosteal bone [55]. Parosteal osteosarcoma originates outside the bone cortex, from the outer layer of periosteum and radiographically is attached by a broad base stalk segregated from the underlying cortex by a slim radiolucent rim. Parosteal osteosarcomas can grow to encircle the underlying bone [60] and extend into the soft parts without clear borders, which are often seen in BPOP, particularly on MRI scans [17].

Histology

Histologically, BPOP is made of three different structures: bone, cartilage and spindle cells. Typical microscopic findings include: 1. disorganized bony trabecula [36] with uniform osteoblasts, 2. spindle-shaped fibroblasts which may have mild atypia [6] located in the intertrabecular space and 3. cartilaginous cap, structured geographically or as irregular islands with atypical, enlarged binucleate (bizarre) chondrocytes [36, 61]. A characteristic feature is the blue color of calcified cartilage on hematoxylin and eosin stains, also called „blue bone" [9].

Florid reactive periostitis is characterized by spindle cell proliferation with absent or less cartilage and with the membranous type of ossification while BPOP demonstrates endochondral ossification. Turret exostosis is composed of cancellous bone with a cortical surface [55]. There is no evidence of a cartilage cap, a clear finding in BPOP. However, many literature cases describe turret exostosis as an osteocartilaginous lesion, while the original work by Wissinger et al. does not describe the presence of a cartilage cap [55]. Theory, first published by Yuen et al. [62], proposes the existence of a continuum among florid reactive periostitis, BPOP and turret exostosis while Kapukaya et al. [51], do not agree with the view that BPOP lesion represents an intermediate stage because their study described four stages of BPOP and each stage carried the histopathological characteristic of BPOP.

A typical characteristic of parosteal osteosarcoma is great difficulty in histological reading in the absence of clear signs of malignancy. Microscopic examination demonstrates parallel, well-formed bony trabeculae in a hypocellular stroma with or without osteo-

blastic rimming and therefore many cases of low-grade parosteal osteosarcoma are considered benign lesions [63].

Management

En bloc excision of the lesion with its pseudocapsule, underlying periosteum and suspicious cortex is the treatment of choice in the first decision for Nora`s lesion [9,64]. Wide resection is the second choice and sometimes amputation is required. Tendency to recur is about 29-55% [1, 6]. In our review (Tab. 1), we were able to detect 74 cases, of which 35 (47.3%) recorded a recurrence. After excision, follow-up of patients every six months is recommended in the first year and then yearly for the next two years [25].

CONCLUSION

BPOP of the hand is a benign lesion, which sometimes behaves aggressively with rapid growth and high risk of local recurrence after resection. BPOP shares many similarities with a spectrum of lesions of which osteochondroma is most common. In our case, we described a typical presentation of BPOP confused with an ostoechondromatous lesion due to its rareness. Medullary continuity of a BPOP lesion of the hand is very rare as we found only one representative case of the 184 reported. BPOP lesions should be managed by an interdisciplinary team to avoid misdiagnosis while histologic examination seems to be the most reliable method.

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