

### Hand's aneurysmal bone cyst: A rare localization. Case report and systematic literature review

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#### **Abstract**

Aneurysmal bone cysts are benign, rare bony tumours frequently observed among children and young adults principally located in the long bones, pelvis, and spine and rarely in other anatomical district such as the hand. We report the case of a 12-year-old girl with an aneurysmal bone cyst, in active stage, involving the still-open epiphysis of the fourth metacarpal of the right hand, which was in a first time treated by curettage, and 3 months later, occurring a recurrence, by a radically excision of the bone and reconstruction with a graft from the iliac crest. At 10-year follow-up the patient had good cosmetic results and a functioning hand. We also performed a systematic Literature review in order to retrieve the key information regarding: the diagnosis, the clinical features and the treatment.

#### Introduction

Aneurysmal bone cysts (ABC) are benign, rare bony tumours that constitute only 1-2% of all bone tumours, described firstly by Jaffe and Lichtenstein in 1942.1 ABC can be primary or can arise from a preexistent lesion however the etiology remains unknown. ABC are most common in the youth, principally located in the long bones, without epiphysis involvement, pelvis and spine. The localization in other anatomical district such as the hand.2 Histologically, ABC appear as multicystic, lytic lesion with cavernous spaces stuffed with blood. The walls of cysts contain fibroblasts and thin strips of bone. The tumors are separated from the surrounding tissue by a thin layer of periosteal new bone.3 The most common treatment of an aneurysmal bone cyst is surgical curettage of the lesion, sometimes filling of the cavity with a bone graft and intraoperative adjuvant therapy may be required. Usually the prognosis following treatment is satisfactory. However, a recurrence rate was reported in the first 2 years after treatment from 10 to 59%, especially in young patients due to skeletal immaturity.4 The recurrence rate also depends on the histopathological pattern of the lesion. Preoperative staging and stagedependent surgical procedures are essential for treatment of ABC and the risk of local recurrence is linked to aggressiveness of the primary lesion and to efficacy of the surgery. The aim of the study is to describe a case of a patient with IV metacarpal bon ABC. We also performed a systematic review of the literature in order to retrieve the key information regarding: the diagnosis, the clinical features and the treatment.

#### **Material and Methods**

#### Search Criteria

The study was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Figure 1). A systematic review of the literature indexed in PubMed, MEDLINE, Cochrane Library and Scopus databases, using as search-terms "Aneurysmal", "Aneurysmatic", "bone", "cyst", "hand" and their MeSH terms combinations (using Boolean operator AND, OR) was performed from 1950 to March 2020. The research was repeated until March 6, 2020.

#### **Inclusion and Exclusion Criteria**

The inclusion criteria of the review were the presence in the evaluated manuscript of: demographic features, symptoms, diagnostic settings, treatment, possible complications and outcomes in patients with ABC of the hand. Only article written in English and available abstract were included. Were excluded from the review: surgical technique reports, expert opinions, studies on animals, unpublished reports, cadaver or in vitro investigations, book chapters, abstracts from scientific meetings.

#### **Data Collection**

Two independent reviewers (A.P and R.V.) separately conducted the described search by title and abstract. If the articles met inclusion criteria following a title and abstract screened, the full text was obtained and reviewed. Any discordance was solved by consensus with a third author (R.D.V.). From each included article were extracted: age and gender of the patients, location of the ABC, type of surgical treatment performed, risk factors, complications related to the treatment performed and

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Informed consent: Written informed consent for scientific purposes and clinical data collection was obtained according to institutional protocol.

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duration of follow-up. Numbers software (Apple Inc., Cupertino, CA) was used to tabulate the obtained data.

### **Statistical Analysis**

Categorical variables are presented as frequency and percentages. Continuous variables are presented as means and standard deviation.

#### Case Report

A 12-year-old girl, with history of pain in her right hand since 2 months, was visited





at our Emergency Unit. On physical examination there was a slight swelling in the dorsal region of the hand, painful to acupressure. The performed radiographs showed a cystic lesion with expanded cortical near the distal region of the fourth metacarpal, close to the still-open epiphysis (Figure 1a). A MRI was requested. Meanwhile the patient was treated in another hospital by simple surgical curettage. In following three months, she had pain and worsening of dorsal hand swelling. The radiographs and MRI showed a cystic lesion with expanded cortical involving all the fourth metacarpal, also the growth plate (Figure 2). The cortical bone was expanded and thin, which made it impossible to remove the tumour by curettage and filling the cavity with bone graft. Instead the entire diaphysis, including the tumour, was removed and a 7 cm bicortical iliac crest graft, oversizing metacarpal dimension, was harvested and fixed with Kirschner wires to the proximal phalanx and then left in place for six weeks (Figure 3). The hand was also immobilized in a short arm cast for the first 4 weeks. Follow-up radiographs showed that the bone graft healed in the correct position. Macroscopical pattern and microscopy of the tumour showed the typical features of the aneurysmal bone cyst with thin strips of bone and fibroblasts surrounded by blood vessels. At ten year follow up range of movement was satisfactory (Figure 4) only the strength in the operated hand was less than in the other hand but she didn't have functional limitation and referred a normal

60 cases (72.3%), pathologic fracture of the involved bone in one case (1.2%). In 47 cases (56.4%) pain and swelling were associated.

Hand's ABC seems to be prevalent in the metacarpal bones (47 patients, 56.4%), followed by the proximal phalanx (19 patients, 23.8%), the middle phalanx (6 patients, 7.2%), the capitate (3 patients 3.6%), the distal phalanx (3 patients, 3.6%), the lunate (2 cases, 2.4%), the hamate (in one

case 1.2%), and the trapezium (in one case 1.2%). In one case the ABC was localized in a sesamoid bone of proximal interphalangeal joint of the index. In 14 patients (16.8%) a traumatic injury of the affected hand was reported. In all case except one (98.8%) a histological diagnosis was made.

# Treatment and outcomes and complication

Eighty-two patients (98.8%) were

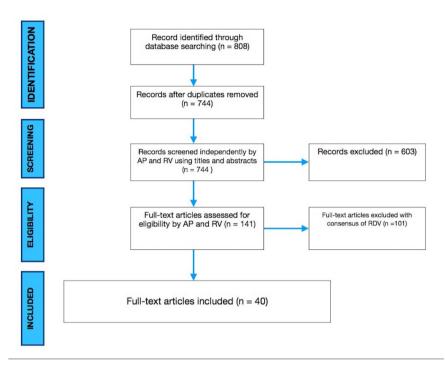


Figure 1. The PRISMA flow-chart.

#### Systematic literature review

## Patients features and demographical data

life. The radiographs are shown in Figure 4.

Only a few cases of hand's ABC were reported in the Literature. A total of 744 reports, excluding duplicates, were independently screened, thereby 40 were finally included in our review (Figure 1). Our review showed 83 cases, including our patients, of hand's ABC in the Literature. (4-55) The mean age of included patients was 18.6 (+/- 10.6) years; 50 patients (60%) were paediatric (< of 18 years) the Male/Female ratio was 1.1; the mean follows up time was 52.1 months (+/-52.2). Demographic and clinical features are summarized in Table 1.1-51

# Symptoms onset, localization, risk factors and diagnosis

All patients had an onset symptom. Presentation symptoms were: hand's swelling in 70 patients (84%), hand's pain in

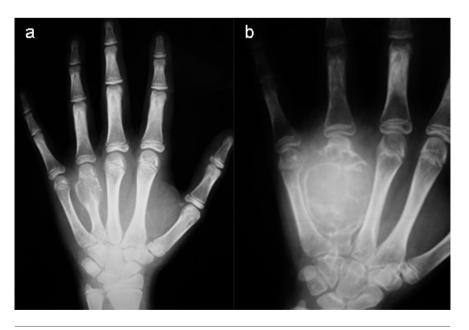


Figure 2. a) Rx pattern before simple curettage. b) Rx pattern three month later simple curettage.



surgically treated. The chosen surgical approaches were: tumour resection and autologous bone graft in 37 cases (44.4%), curettage of the lesion and autologous bone graft in 26 cases (31.2%), curettage of the cyst in 6 cases (7.2%), curettage and bipolar cauterization in 6 cases (7.2%), tumour excision in 6 cases (7.2%), amputation in 2 patients (2.4%) and conservative treatment only in one case.

Concerning surgical outcome: in 68 patients (81.6%) the first surgery was curative without signs of recurrence. Recurrence of the disease was found in 15 patients (18%), therefore they underwent reoperation. In 8 of this patients, tumour resection and autologous bone graft was used as rescue surgery. In 4 patients after recurrence a new curettage of the lesion and autologous bone graft was performed. In one patient after recurrence, cryotherapy and curettage was used as rescue surgery. In one patient the reoperation consisted in the amputation of affected finger. In one patient the reoperation consisted in curettage and bipolar cauterization of the lesion.

About complication, in 15 patients (18%) was found a limitation of range of motion (ROM); therefore, in 5 of these patients was necessary surgical debridement and tenolysis. In 3 paediatric patients (3.6%) a premature physeal closure was found.

#### Discussion

The origin of the term "aneurysmal bone cyst" derived from two cases of unicameral bone cysts reported by Jaffe and Lichtenstein in 1942. In that report, they noted two large "peculiar blood-containing cysts," which they described as "aneurysmal bone cyst". Jaffe argued that aneurysmal bone cysts could be the result of an hemorrhagic "blowout" in a preexisting lesion, which may be destroyed in the process.1,2 Lichtenstein instead proposed a vascular origin, without specifying whether this lesion was a localized venous thrombosis or an congenital arteriovenous malformation.3,4 Although many hypotheses have been developed over the years, today the nature of ABC is unclear. Many authors defined aneurysmal bone cysts as a secondary evolution of a pre-existing lesion.<sup>2,3</sup> Other authors proposed two different aetiologies characterizing the lesion as either primary or secondary to a known precursor.45 Most cases are found among children and young adults, in fact the majority of patients with aneurysmal bone cysts are younger than age 20 years. These lesions are principally located in the long bones, pelvis, and spine. Most rarely it is

observed in the hand. 4,19,22

The natural history of ABC is characterized by four radiologic stages: initial, active, stabilization, and healing. In the initial phase, the lesion is composed of a well-defined area of osteolysis. During the growth phase the lesion grows exponentially leading to the "destruction" of the bone and to the typical "blown-out" radiological

appearance. Then follows a period of stabilization defined on the X-ray as having a "soap bubble appearance" (which is caused by the maturation of the bony shell). Final healing results in progressive calcification and ossification, with the lesion transformed into a dense bony mass.<sup>46</sup>

There has been no agreement on a definitive or ideal treatment in the entire



Figure 3. Rx pattern of reconstruction.

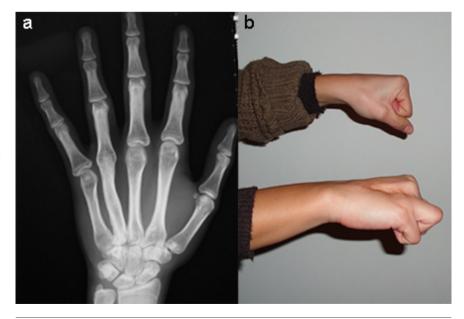


Figure 4. a) Rx at ten years follow up. b) Flexion of fingers at ten years follow up.





Table 1. Demographic and clinical features.

Study	Case	Sex	Age	Risk	Symptoms	Localization	Complication	Surgical	Donor	Outcome	Follow
			(Year	·) Factors							Jp (montl
Mason <i>et al.</i> . 1958	1	M	9		P, Sw	PP III F		CABG	Iliac crest	Cured	33
Harto-Garofalidis <i>et al.</i> 19	967 2	M	17		P, Sw	PP I F	Limitation of ROM	TRABG	Tibia	Cured	
Chari <i>et al</i> . 1971	3	F	16		P, Sw	IV MTB		TRABG	Tibia	Cured	10
Burkhalter <i>et al.</i> 1978	4	F	22	_	P, Sw	IV MTB	Limitation of ROM	TRABG	Iliac crest	Cured	12
Burmantor et al. 1970	5	M	8	Trauma	P, Sw	IV MTB	Limitation of ROM	TRABG	Fibula	Cured	18
	6	M	10		P, Sw	III MTB Lim	nitation of ROM, Recurrence	1° Curettage 2° TRABG	Fibula	Reoperation/Cured	72
Fuhs <i>et al</i> . 1979	7	M	17	Trauma	P, Sw	DP IV F		Amputation	-	Amputation	12
	8	M	20	Trauma	P, Sw	I MTB	Limitation of ROM	TRABG	Iliac crest	Cured	-
Chalmers et al. 1981	9 10	M F	36 13	-	P, Sw P, Sw	PP V F PP II F	Recurrence	CABG CABG	-	Reoperation/Cured Cured	-
	11	M	16	-	P, Sw	PPIF	_	Curettage	_	Cured	_
Barbieri <i>et al.</i> 1984	12	F	35	_	P, Sw	IV MTB	Limitation of ROM	F trasposition		Cured	24
	13	F	11	_	P, Sw	II MTB	_	TRABG	_	Cured	48
	14	F	30	-	P, Sw	IV MTB	-	CABG	-	Cured	6
Lin <i>et al.</i> 1984	15	M	16	Trauma	P, Sw	Hamate	_	TE	_	Cured	15
Frassica et al. 1988	16 17	F M	13 20	-	P, Sw	PP V F	Recurrence	CABG TE	-	Reoperation/Cured Cured	244 176
	18	F	49	_	P, Sw P	Trapezium PP I F	Recurrence	CABG	_	Reoperation/Cured	254
	19	F	36	_	P, Sw	V MTB	_	TRABG	_	Cured	132
	20	M	28	_	P, Sw	III MTB		CABG	-	Cured	55
	21 22	M M	55 14	-	P PF	DP V F I MTB	Recurrence	CABG Curettage	-	Cured Reoperation/Amputation	59 42
	23	F	14	_	P	IV MTB	_	CABG	_	Cured	43
	24	F	28	_	P	I MTB	D	CABG	-	Cured	32
V . 1 . 1 1000	25	F	16		P	V MTB	Recurrence	1° CABG 2° TRABG	— —	Reoperation/Cured	27
Kotwal et al. 1988	26	F	5	Previous surgery CABC		III MTB	_	TRABG	Fibula	Cured	48
Kozlowski <i>et al</i> . 1988	27 28	M M	8 9	Trauma	P, Sw P, Sw	III MTB DP III F	-	CABG Curettage	-	-	-
	29	F	10	Trauma	P, Sw	II MTB	_	TRABG	_	_	_
	30	M	11	_	P, Sw	MP III F	_	CABG	_	_	-
	31	F	13	-	P, Sw	II MTB	-	CABG	-	-	_
Milliez et al. 1988	32	F	10	m	P, Sw	MP III F	Recurrence	CABG	Iliac crest	Reoperation/Cured	6
Dossing et al. 1990	33	F	40	Trauma	P, Sw	MPIF	Recurrence	1° CABG 2° TRABG	_	Reoperation/Cured	18
Mortensen <i>et al.</i> 1990	34	M	6	_	P, Sw	PP III F		TRABG	Ulna	Cured	72
Rao <i>et al.</i> 1993	35 36	M F	31 11	-	P c	I MTB MP II F	Recurrence	1° CABG 2° TRABG 1° CABG 2° TRABG	IV MTB	Reoperation/Cured	44 6
Borrelli <i>et al</i> . 1994			10	_	Sw	III MTB	Recurrence	TRABG			30
	37	M	40	_	Sw		Limitation of DOM		Fibula	Cured	
Mankin et al. 1995	38	F		—	P	Lunate	Limitation of ROM	TE, SCA	_	Cured	6
Platt et al. 1995	39	F	14	Trauma	P P	Capitate	_	No TDADG		Cured	16
Apaydin et al. 1996	40	M	32	_ m	P, Sw	I MTB	-	TRABG	Iliac crest	Cured	18
Sakka et al. 1997	41	M	8	Trauma	P, Sw	DPIF		Curettage	_	Cured	36
Athanasian <i>et al.</i> 1999	42	F	14	Trauma	P, Sw	PP III F	Recurrence	1° CABG 2° Criosurgery	Iliac crest	Reoperation/Cured	60
Sproule et al. 2002	43	M	8	Trauma	P, Sw	PPIF	Limitation of ROM	CABG	_	Cured	12
Gundes et al. 2005	44	M	25	_	P, Sw	IV MTB	Limitation of ROM	TRABG	Fibula	Cured	12
Havulinna <i>et al.</i> 2005	45	M	34	_	Sw	Sesamoid II F	_	TE		Cured	24
Basarir et al. 2006	46	M F	18 22	-	P, Sw	V MTB PP IV F	– Pogurrongo	CABG 1° CABG 2° TRABG	Iliac crest	Cured Reoperation/Cured	18 22
	47 48	и	16	_ Trauma	P, Sw P, Sw	IV MTB	Recurrence	TRABG	Iliac crest Iliac crest	Cured	14
	49	F	52	Trauma	P, Sw	II MTB	Recurrence	1° CABG 2° TRABG	Iliac crest	Reoperation/Cured	125
Sakamoto <i>et al.</i> 2006	50	M	15		P	Capitate		Curettage		Cured	48
Sahu <i>et al</i> . 2008	51	F	12	_	Sw	I MTB	_	TE	_	Cured	24
Ozyurek <i>et al.</i> 2009	52	M	21		P, Sw	V MTB		TRABG	Iliac crest	Cured	36
Jafari <i>et al</i> . 2011	53	M	16	_	Sw	I MTB	Limitation of ROM	TRABG	Fibula	Cured	
	54	F	6	_	Sw	III MTB	_	TRABG	Fibula	Cured	_
	55 Ec	M	17	-	Sw	I MTB	Croft recombine	TRABG	Fibula	Cured	-
	56 57	M F	27 15	-	Sw Sw	PP V F III MTB	Graft resorption	TRABG TRABG	Iliac crest Iliac crest	Cured Cured	-
	58	F	10	-	Sw	II MTB	_	TRABG	Fibula	Cured	_
	59	M	9	_	Sw	V MTB	_	TRABG	Fibula	Cured	_
	60	F	23	_	Sw	II MTB	Recurrence	TRABG	Iliac crest	Reoperation/Cured	18
	61 62	F F	15 16	_	Sw Sw	V MTB	Limitation of ROM Limitation of ROM	TRABG TRABG	Fibula Fibula	Cured Cured	-
	62 63	r M	17	-	Sw Sw	III MTB PP II F	MINIMUNII OI NOM	TRABG	Fibula Iliac crest	Cured	-
	64	F	20	-	Sw	PP IV F	_	TRABG	Iliac crest	Cured	-

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Table 1. Continued from previous page.

Study	Case	Sex	Age (Year)	Risk Factors	Symptoms	Localization	Complication	Surgical treatment	Donor site	Outcome	Follow Up (month)
Moussallem et al. 2011	65	M	20	_	P	Capitate	_	CABG	Iliac crest	Cured	24
Kieseritzky <i>et al.</i> 2012	66	M	15	Trauma	P, Sw	III MTB	_	TRABG	Radius	Cured	144
Rajappa et al. 2013	67	F	23	_	P	Lunate	_	TE	_	Cured	72
Singh <i>et al.</i> 2013	68	M	6	_	Sw	III MTB	_	TRABG	Fibula	Cured	24
Al-qattan et al. 2014	69	M	11	_	Sw	MP III F	Asimmetry, PPC	CC	_	Cured	36
	70	F	12	_	Sw	IV MTB	PPC	CC	_	Cured	36
	71	M	6	_	Sw	PP IV FRecu	rrence, PPC, Limitation of ROM	1°CABG 2°CC	_	Reoperation/Cured	36
	72	F	20	_	Sw	PP III F	_	CC	_	Cured	36
	73	M	28	_	Sw	MP V F	_	CC	_	Cured	36
	74	F	22	_	Sw	PP IV F	_	CC	_	Cured	36
Pallapati <i>et al</i> . 2016	75	M	29	_	P, Sw	V MTB	_	TRABG	VI MTB	Cured	85
	76	F	14	_	P, Sw	III MTB	_	TRABG	VI MTB	Cured	84
	77	F	15	_	P, Sw	III MTB	_	TRABG	IV MTB	Cured	84
	78	M	23	_	P, Sw	III MTB	_	TRABG	III MTB	Cured	90
Zancolli <i>et al.</i> 2016	79	M	19	_	P	Pisiform	_	TE	_	Cured	_
Nanda <i>et al</i> . 2018	80	F	12	_	P, Sw	III MTB	_	CABG	Iliac crest	Cured	6'
Zaidenberg et al. 2018	81	M	21	_	P, Sw	IV MTB	Limitation of ROM	TR Bone Allograft	IV MTB	Cured	96
Dhamangaonkr et al. 2020	82	M	2	_	P, Sw	PP III F	_	CABG	_	Cured	60
Present case	83	F	12	_	P, Sw	IV MTB	Limitation of ROM	1° Curettage 2° TRABG	Iliac crest	Reoperation/ Cured	120

CABG: Curettage and autologous bone graft; CC: Curettage and cauterization; Dp: distal phalanx; P: finger; MCB: metacarpal bone; MP: middle phalanx; MTB Metatarsal bone; P: pain; PF: pathologic fracture; PP: proximal phalanx; PPC: premature physeal closure; ROM: Range of Motion; SCA: scapho-capitate arthrodesis; Sw: swelling. TE: tumor excision; TRABG: Tumor resection, curettage and autologous bone graft.

scientific literature which is why many different treatment options are used.

Conventional treatments of an ABC, partly similar to simple cysts treatment, <sup>47,48</sup> were represented by: the surgical removal of the entire lesion (en-block or piece-meal) or curettage of the lesion, with or without bone graft, application of liquid nitrogen or by reconstruction using an intramedullary rod, bone auto-graft, polymethylmethacrylate with Steinmann pins, Masquelet technique, implantation of allograft chips. <sup>5-44,49</sup> The efficacy of these methods, specifically of intramedullary nailing and bicortical or tricortical bone grafts from the iliac crest, had already been demonstrated in other studies. <sup>50,51</sup>

Due to its rarity in the hand, no evidence-based treatment regimen has been established, however, from our systematic review it emerged that a radical treatment with tumour resection and bone autograft was associated with the best result and with the lowest number of recurrences. This treatment can be used also as a rescue treatment in the event of disease recurrence.

Our case shows that good results can be achieved although the tumour involved the growth plate and had destroyed the entire diaphysis of the metacarpal and also shows that a bicortical iliac crest graft can transform into an almost normal-looking bone.

#### **Conclusions**

Although most authors recommend a less radical approach, a wide resection and a cortical bone graft is indicated in cases when the articular surface or growth plate is involved or when full bone invasion occurred.

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