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Case Report

Skeletal manifestations of hydatid cystic disease in Sudan

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الملخص

أهداف البحث: مرض الكيس العداري العظمي هو مرض حيواني يسببه في الغالب "اكينوكوكس جرانيولوسس". ويُعتبر سكان دول الشرق الأوسط وأفريقيا والبحر المتوسط معرضين للإصابة بصورة وباء محلي. يُعد مرض الكيس العداري في العظام حالة سريرية نادرة تصيب الذين لديهم تاريخ من الاتصال بالحيوانات. يهدف هذا البحث إلى مراجعة الأعراض السريرية للتكيس العداري في الجهاز العظمي.

طرق البحث: تم جمع بيانات جميع الحالات السريرية المصابة بالكيس العداري في الجهاز العظمي خلال الفترة ٢٠١١ - ٢٠١٧. كما تمت مراجعة الأعراض السريرية وتقارير الأشعة والأنسجة، وتمت متابعة المرضى لمدة ١٢- ٢٢ شهرا.

النتائج: أصيب مريضان بمرض العداري في العمود الفقري ومريض في عظم الحوض ومريض في عظمة القص ومريض في عظمة الفخذ. وكانت الأعراض توحي بوجود كسور مرضية وتشوهات عظمية. صاحب مرض العداري في العمود الفقري ضغط على جذر العصب وضعف النصف السفلي للجسم في حالة، وألم جذري في الحالة الأخرى. وظهر مع إصابة عظمة القص ألم وتشوه، في حين صاحب إصابة عظمة الفخذ تشوه وكسر. تم الاستئصال الجراحي مع استخدام دورة مضاد (طارد) الديدان قبل وبعد العملية وتطبيق اسلوب اختراق الكيس ثم الرشف ثم حقنه ثم الرشف في أربع حالات. وأظهرت حالة واحدة وجود مرض متبقي خلال زيارات المتابعة.

الاستنتاجات: يُعتبر مرض الكيس العداري في العظام صورة نادرة لمرض منتشر. ويُعد النهج متعدد التخصصات هو الأسلوب الأمثل للتعامل مع هذه الحالات. المتابعة طويلة المدى إلزامية للحماية من المضاعفات ومعاودة المرض ومراقبة الإعاقات المتبقية.

الكلمات المفتاحية: مرض العداري؛ اكينوكوكس؛ جرانيولوسس؛ حيواني المنشأ؛ مرض العداري في العمود الفقري

Abstract

Objectives: Hydatid bone disease is a zoonotic disease caused mainly by *Echinococcus granulosus*. Middle Eastern, African, and Mediterranean populations are endemically affected. Hydatid disease of the bone is a rare clinical condition that affects people with a history of animal contact. This research aims to revisit clinical presentations of hydatid cystic disease in the skeletal system.

Methods: Data of all clinical cases with hydatid cystic disease of the skeletal system, presenting between 2011 and 2016 were collected. Clinical manifestations and radiographic and histopathological reports were reviewed, and the patients were followed for a period of 12–22 months.

Results: Two patients had hydatid disease in the spine, one in the pelvis, one in the sternum, and one in the femur. The symptoms were suggestive of pathological fractures and bony deformities. Spinal hydatid disease presented with nerve root compression and paraparesis in one case, and radicular pain in the other case. Sternal involvement of hydatid disease presented with pain and deformity, while femoral involvement presented with deformity and fracture. Surgical resection with a pre- and post-surgical anthelminthic course and application of the puncture, aspiration, injection, and aspiration (PAIR) technique were completed in four cases. One case showed residual disease during follow-up visits.

Conclusions: Hydatid cystic disease of the skeletal system is a rare presentation of a common disease. A multidisciplinary approach is optimal for the management of these cases. Long-term follow-up is mandatory to prevent complications and recurrence, and to monitor residual disabilities.

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Keywords: Echinococcus; Granulosus; Hydatid; Hydatid disease spine; Zoonosis

Introduction

Hydatid cystic disease is a zoonotic disease caused by the Echinococcus sp. tapeworm. The definitive hosts are carnivores including dogs, wolves, and foxes; and the intermediate hosts are herbivores including cattle, sheep, and deer. Humans act as accidental dead-end hosts.

There are four recognized species within the genus Echinococcus (E.): *E. granulosus* causes hydatid cystic disease and 10 different genotypes have been described, *E. multilocularis* causes hydatid alveolar disease, *E. vogeli* causes hydatid polycystic disease, and *E. oligarthus* causes polycystic disease.[1]

Echinococcus granulosus hydatidosis is found in cropping areas, including agricultural, cattle, and sheep rearing communities. The dual existence of herbivores and carnivores enables the cycle to be completed, accompanied by inadvertent transmission to humans in close proximity.

African and Middle-Eastern countries are considered endemic areas. Sudan is a vast country with immense landscapes and cattle and sheep livestock populations. Hydatid cystic disease of different organs is habitually detected in different pathological specimens with a myriad of clinical presentations.

The life cycle in the definitive carnivore host starts by invasion of the protoscolex and attachment of the scolex to the wall of the intestine. This matures to an adult worm composed of three proglottids. The terminal proglottid is the gravid uterus, which lays embryonated eggs in dog faeces.

After accidental ingestion of contaminated food, the life cycle is completed in the herbivore intermediate host, or the accidental host (humans), with the hatching of the embryonated eggs to produce an oncosphere. This penetrates the intestinal wall and gains access to the portal circulation, and subsequently to the systemic circulation.

The circulating oncosphere resides in different tissue sites, preferably, in descending order: the liver, lungs, spleen, kidney, heart, bone, and the central nervous system. The cycle is completed when the carnivores eat the remains of the infected herbivores.

The signs and symptoms vary according to the afflicted site. These are mainly attributed to the slowly expanding cyst inducing pressure within the organ, causing atrophy or obstruction of ducts or blood vessels. Rupture of the cyst with the liberation of the intensely immunogenic materials can cause catastrophic anaphylactic shock. Skeletal lesions of the axial skeleton can result in deformities, pathological fractures or collapse, possibly causing paraplegia or root compression signs and symptoms. Appendicular skeletal lesions present with deformities and pathological fractures.[2]

Materials and Methods

All cases presented over a 5-year period from 2011 to 2016. The patient's clinical data and radiographic and histopathological reports were reviewed, and further information was obtained by contacting the patients or their relatives. Informed consent was granted from the enrolled patients to anonymously use their clinical data.

Personal and clinical data (history and examination), radiographic and surgical reports, and a detailed gross and microscopic description of the provided specimen, were obtained and revised.

Biopsies were obtained to exclude other possibilities prior to formal excision, including unicameral bone cysts, or cystic degeneration of primary or secondary bone tumours.

The PubMed case reports of hydatid cystic disease of the skeletal system were reviewed over the 5-year period. A statistical analysis of our cases was performed and compared against the published reports.

Case reports

Case-1

A 53-year-old female, mother of eight children, complained of vague pelvic pain at night, which extended to the groin. This had persisted for 2 months, and there were no constitutional symptoms or history of trauma. During history taking, we discovered the presence of animals, including three dogs, in her household.

Plain x-ray showed a well-defined cystic lesion with sclerotic margins in the iliac crest. A trephine biopsy, composed of a blood clot and firm tissue, was taken and sent in three blocks for histopathological processing and examination.

The microscopic evaluation of the trephine bone biopsy showed marked histiocytic and giant cell reactions around fragments of acellular laminated hydatid membrane. Viable hydatid scolices were noted. These features are consistent with those of a hydatid cyst of the pelvic bone.

Based on the histopathological diagnosis, the patient underwent surgical resection with a wide margin, preceded and followed by albendazole anthelminthic therapy. The accessible site of the lesion facilitated complete removal. The Puncture-Aspiration-Injection-Reaspiration (PAIR) technique was applied to the lesion to reduce the potential risk of the cyst contents leaking. The patients did not show any clinical or radiographic evidence of recurrence at the 20-month follow-up.

Case-2

A 30-year-old male from Kurdufan presented with central bulging superficial chest pain and swelling. Plain x-rays showed no pulmonary lesions, which excluded the possibility of direct extension of lung cysts. Other visceral or skeletal lesions were excluded during the investigations. Preoperative chemotherapy was undertaken, and the PAIR technique was conducted during surgery with the introduction of hypertonic saline. An excisional biopsy was taken, which was composed of multiple pieces of grey-white tissue containing two thin-walled cysts; the larger cyst was 2 cm in diameter.

The cysts contained clear fluid, and the specimen was sent in one block (Figure 1)

The microscopic evaluation of the cysts showed a strongly stained PAS-positive laminated hydatid membrane lined by a germinal layer with multiple brood cysts and scolices. Wet preparations from the cyst fluid showed multiple hydatid brood capsules, scolices, and hooklets. These features are consistent with those of a hydatid cyst of the sternum (Figure 2).

The patient made a complete recovery, with no residual lesions in the operative or other sites during any of the follow-up visits up to 16 months.

Case-3

A 56-year old male from a small village beside the Blue Nile presented with lower spastic paraparesis, which was more apparent on the right side. Magnetic resonance imaging showed a clear space with the occupying lesion involving the body of the third lumbar vertebra (the report was provided without pictures).

Ultrasonic examination of the liver revealed a small subcapsular parenchymal cystic lesion measuring 2.5 cm, consistent with a hydatid liver cyst. The mass and the surrounding fragments of the bone and soft tissue were taken for histopathology. The specimen was composed of multiple fragments of the bone taken all in one block with decalcification. The accompanying soft tissue was sent in three blocks.

The microscopic evaluation of the specimen showed bone, adipose, and skeletal muscle tissue. The bone was partly replaced by a well-demarcated amorphous eosinophilic material with pale staining well-formed strands embedded in the matrix. The strands were PAS-positive (Figure 3). The bone marrow within the normal fragments of bone showed normal cellularity and no evidence of an increase in plasma cells. No evidence of primary or secondary malignancy was observed in the sections of the biopsy that were submitted. Although the features were suggestive of localised degenerative bone disease, a sterile calcified hydatid cyst was also considered. The final diagnosis after revision of the clinical and ultrasonic data was D3 Vertebra: fibrotic lesion/calcified hydatid cyst.

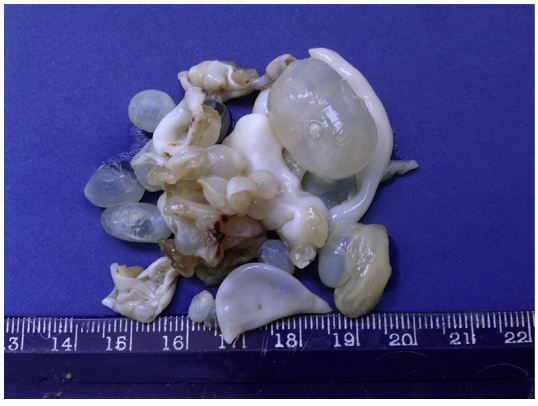

Figure 1: Opened primary *E. granulosus* cyst with multiple daughter cysts carpeting its luminal border. Some cysts are intact and others are flaccid.

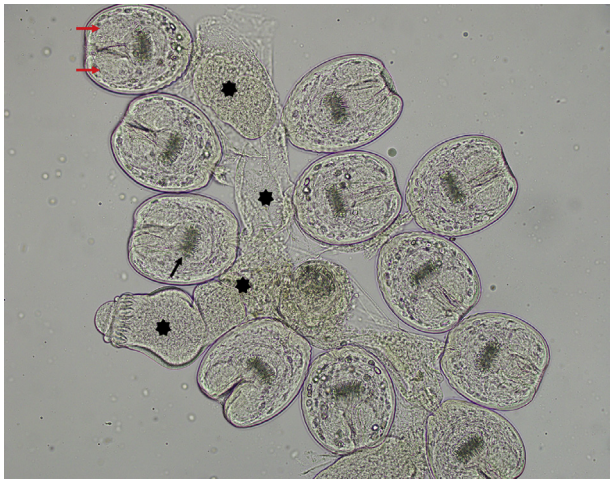

Figure 2: Wet preparation of the cyst content shows hydatid sands of protoscolices, it measures about 100 mm in diameter, and has one to two pairs of suckers (**red arrows**) and rostellum with dual row of hooklets (**black arrows**). In the middle of the field notice unretracted adult worm with gravid terminal bulbous proglottid traced to the top of the field (*).

Our patient showed residual neurological deficits after surgery, with minor but noticeable improvement after 2 months of physiotherapy. No residual lesions were identified in the spine or the liver at the first (1 month) follow-up visit; however, the patient showed poor compliance with the postoperative albendazole treatment. During the second (3 month) follow-up visit, the patient showed conspicuous recurrence of the lesion in radiographic computed tomography scans. Surgical revision of the lesion was undertaken 1 month later (4 months after the initial surgery), and 22 months have elapsed since the last surgery without any recurrence.

Case-4

A 30-year old male working in an abattoir presented with a destructive lesion of the tenth thoracic vertebra (T10), with fluid collection ventral and distal to the cord inducing nerve root compression with radicular pain. During the surgical laminectomy, the spinal cord was released and the lesion was removed. The intraoperative findings included swelling of the spinal cord, and a cavity within T10 filled with a mass encapsulated by tough fibrous tissue. A biopsy was taken from the capsule and the lesion. One container with multiple pieces of bone, measuring 3.0 × 3.0 × 0.3 cm in total, was sent for processing in two blocks. A second container, with multiple pieces of gelatinous white soft tissue measuring 3.0 × 2.0 × 0.3 cm in total, was sent for processing in one block.

The microscopic evaluation of the dorsal spine biopsy showed a laminated acellular membrane. No scolices were identified. These features are consistent with those of a hydatid cyst of the dorsal spine (Figure 3 A, B, C)

The patient received preoperative and postoperative anthelminthic albendazole treatment. The PAIR technique was not introduced during surgery. The patient showed a gradual recovery, and the radicular nerve compression

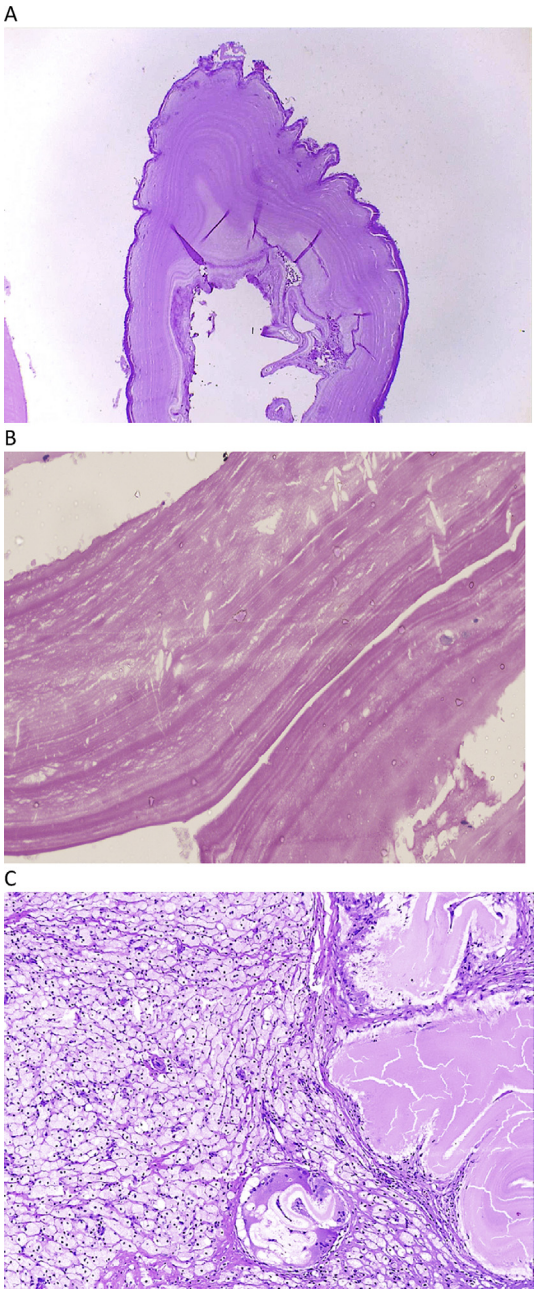

Figure 3: A- a cyst wall of one of the brood corpuscles showing inner germinal and outer laminated membrane. B- The PAS positive laminated membrane. C- Cyst content spillage with a surrounding histiocytic reaction, at the bottom notice a giant cell engulfing part of the membrane.

symptoms improved with physiotherapy. No residual lesions were identified at the 3-, 6-, and 12-month follow-up visits.

Case-5

A 40-year-old male working as a farmer presented with a pathological fracture of the right femur. The provisional diagnoses from the x-ray scan were primary bone tumour or hydatid disease (Figure 4). Surgical resection with the introduction of a fixation device was performed without preoperative albendazole treatment or the PAIR technique.

Bone curettage was performed and three containers were used for the biopsy. The first container of multiple pieces of grey-white soft tissue, measuring 6.0 × 3.0 cm in total, was sent for processing in three blocks. The second container of multiple pieces of soft tissue (cyst wall), measuring 4.0 × 3.5 cm, was sent for processing in two blocks. The third container of multiple pieces of soft tissue and blood clot, measuring 13.5 × 10.0 cm, was sent for processing in five blocks.

The microscopic evaluation of the right femur biopsy samples showed the lamellar membrane surrounded by fibrosis, and marked inflammatory infiltrate composed mainly of giant cells, lymphocytes, and macrophages. The PAS stain yielded positive result. These features are consisted with those of hydatid disease of the right femur. The patient showed no radiographic, clinical, or surgical evidence of recurrence at the follow-up visits or during the removal of the fixation device after 14 months.

Discussion

The medical literature from the Middle East and Africa shows that hydatid bone diseases are under-reported. This

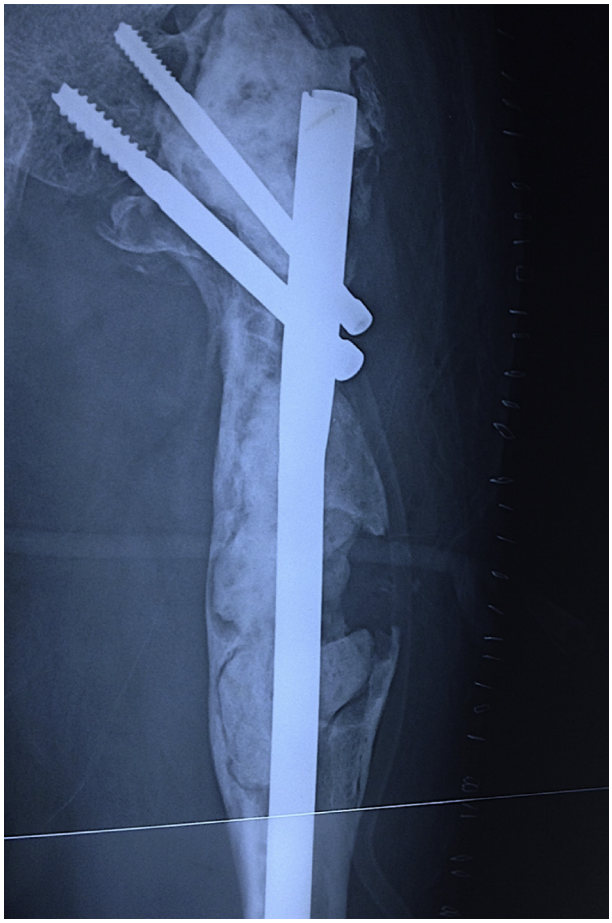

Figure 4: Hydatid disease of the femur showing well defined radiolucent locular lesions extending down to the mid-shaft of the bone. Also note the internal fixation device traversing the site of the pathological fracture.

Table 1: Case reports of hydatid bone disease extracted from PubMed starting from 2012 to 2017.

#	Author (Number of cases/report)	Year	Country	Age (mean)	Gender	Site
1	Alimi	2012	Tunis	40	M	Spine
2	Senol MG	2012	India	55	F	Spine
3	Florence Leslé (2)	2013	France	38	1M + 1F	Femur
4	Kaboor	2013	India	47	M	Femur + Pelvis
5	Aurore Keutgens	2013	Belgium	75	M	Spine
6	Doğanavşargil B (2)	2014	Turkey	39.5	1M, 1F	Pelvis
7	Sayed Walid	2014	Tunis	24	M	Pelvis
8	Nourrisson	2014	France	82	F	Spine
9	Rachid Marouf	2014	Morocco	32	F	Spine
10	Joshi, K. C.	2014	India	28	M	Spine
11	Yashdeep Sarma	2014	India	64	M	Spine
12	Kassa BG	2014	Ethiopia	53	F	Tibia
13	Varela	2015	Chile	50	F	Spine
14	Erol B (10)	2015	Turkey	42.5	6F, 4M	Muscle (8), bone (2)
15	Babitha	2015	India	52	F	Femur
16	Khan MS	2015	Pakistan	30	F	Femur
17	Zengru Xie (40)	2015	China	41.5	24M, 16F	Femur = 2, pelvis + femur = 17, chest = 15, spine = 10
18	Tsagozis	2015	Sweden	57	M	Pelvis
19	Roman A	2015	Romania	25	M	Chest bones
20	Rabiou (3)	2016	Morocco	41	M	Chest bones
21	Gennari	2016	France	25	F	Spine
22	Abbasi	2016	Iran	61	M	Sacrum
23	Serbest	2016	Turkey	22	F	Scaphoid
24	Togral (5)	2017	Turkey	41.6	3M, 2F	Iliac bone (3), Femur (1), thigh (1)
25	El Hammoumi	2017	Morocco	28	M	Spine

undermines the medical and economic impact of these lesions, and decreases the awareness of the general and medical community of its clinical presentation and potential complications.

To our knowledge, these are the first reported cases of hydatid cystic disease of the skeleton in Sudanese patients. In the 5-year period examined, six reports from African countries, one case report from Ethiopia,[3] three from Morocco[4–6] and two from Tunisia,[7,8] were identified in PubMed.

We presented five cases of *E. granulosus* osteomyelitis from Sudan; one involving the pelvic bone, one involving the sternum, two involving the spine, and one involving the femur. Eighty percent of the cases arose within the axial skeleton (40% from the spine, 20% from the chest wall and 20% from the pelvic bone), and 20% arose from the appendicular skeleton (the femur). Eighty-one cases were reported in the medical literature extracted from PubMed in the 5-year period (Table 1). Twenty of the published cases (25%)

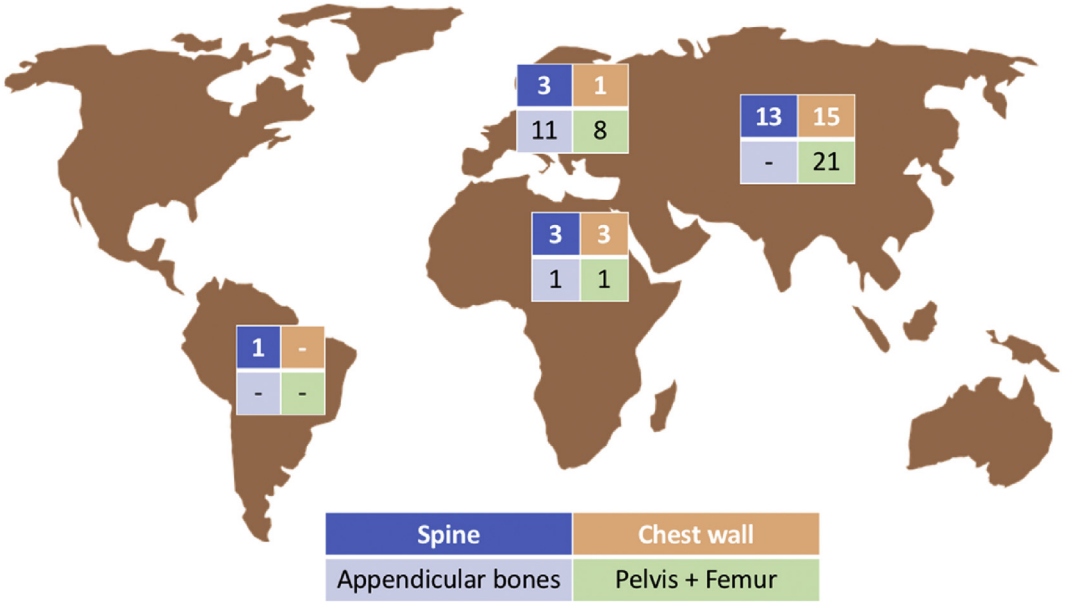

Figure 5: Geographic distribution of hydatid bone disease of the case reports in PubMed from 2012–2017.

affected the spine. Three cases were from Africa (Tunisia, Morocco, and the Mediterranean),[7,6,5] three cases were from Europe,[9–11] 14 cases were from Asia[12–15] and one case was from Chile.[16] Two cases from the PubMed reports were of alveolar echinococcosis caused by E. multilocularis, which showed multiple bubbly cystic bone lesions.[9,10] One case report from Turkey identified this disease in the scaphoid bone, a small carpal bone in the hand. Fortunately, it was discovered due to the high disease prevalence and high index of suspicion.[17]

The appendicular, chest, and pelvic bones were affected in 61 cases (75%) (Figures 5, 6). Thirty-six cases were from Asia; 15 involved the chest wall (19%), and 21 involved the pelvis and adjacent femur (26%).[18–20,15,21] From Africa, one case involving an appendicular bone was reported from Ethiopia (1%), three reported cases affected the chest wall (3%), and one case involved the pelvis (1%).[4,3,8] Twenty cases involving the pelvis, chest wall and appendicular bones were reported across Europe (25%).[22–26,17,27] Eight cases affected the pelvic bones and/or femur (10%) (Figures 5, 6), while 11 cases involved the appendicular skeleton, with eight of these cases reportedly occurring within the skeletal muscles. There was no recurrence after an extended follow-up (64-month) period.[24]

Many of the spinal and chest wall hydatid cysts were presumed to be a result of the progressive extension of pulmonary or hepatic lesions encroaching upon adjacent bony structures. In these cases, primary parenchymal or soft tissue lesions are discernible on radiographic images indicating the epicentre of the lesion. Many cases arising from the bone showed no accompanying lesions or direct expanding lesions with secondary involvement, indicating the primary origin of the bony lesions.[2] All cases within our study showed isolated primary bone lesions, except case-3, which showed an accompanying liver parenchymal cyst. There were no extra-osseous extensions in any of the cases except case-3, where bone fracture and resorption with extension to the paraspinal soft tissue were evident.

All of the cases reflect the occupational hazard encountered by high-risk groups concerned with cattle or sheep breeding. This was reflected both in our study and in the PubMed reported cases. All of the males showed exposure to animal contact in their history and as a consequence of their occupation (the cases included a farmer, a butcher, and two village dwellers). The affected female in our study reported the presence of household animals, including one cow, five goats, 10 hens, and three dogs.

In our study, one case presented in a female (20%), and four cases in males (80%). In the reviewed reports, 44 cases (54%) were found in men and 41 (46%) in females. This bias towards the male gender reflects the social and occupational impact of the disease, with men in Sudan often travelling to distant districts to seek fertile land for herding. Females are usually concerned with herding small groups of animals in their home for domestic use. The gender distribution ratio of our cases cannot be used to infer global gender ratios for this disease due to the statistical limitations inherent in the number of cases evaluated.

In our study, the age of the patients ranged from 30 years to 56 years, with an average age of 38 years. In the reported cases from PubMed, the average age was 44 years (range: 22–82 years).[17,10] This reflects the social and the economic impact of the disease, which shows its predilection for young adults.

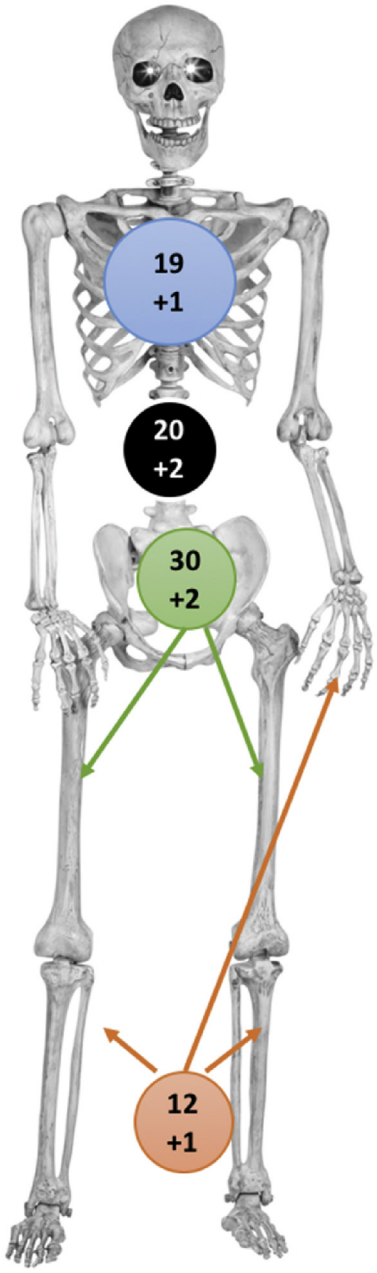

Figure 6: The skeletal distribution according to sites of Hydatid Disease of bone lesions extracted from PubMed case reports from 2012 to 2017 (+ the number of cases in our series).

The PAIR technique, under the guidance of imaging or surgical exposure, with an intralesional installation of a parasiticidal solution or hypertonic saline was applied in all cases, except case-3. A preoperative and postoperative course of oral albendazole anthelminthic therapy, varying in duration from 2 to 4 weeks prior to surgery, was introduced in three cases.

The definitive treatment for our patients consisted of careful surgical shelling or curettage. The pelvic bone, femur, and chest wall lesions were completely cured, with a complete absence of lesions at the 1-, 3-, and 6-month follow-up visits.

The patients with spinal hydatidosis sustained residual neuronal symptoms, with minor but noticeable improvement after physiotherapy.

Careful shelling out or curettage is warranted to safeguard against potential spillage of the cyst contents, which can induce a dramatic anaphylactic reaction. The most important and decisive step in the management of this disease was the feasibility of complete surgical resection. Case-3 showed recurrence of the lesion after a 3-month interval. Despite the preoperative anthelminthic albendazole therapy, the surgeon reported difficulties during the surgical intervention, and the patient reported poor compliance with the postoperative therapy.

In a large meta-analysis, the recurrence rate increased if the bony lesions were associated with visceral involvement; while the duration of the anthelminthic therapy, the number of surgeries, or the introduction of the PAIR technique, had no effect on recurrence risk.[28] Case-3 had accompanying hepatic visceral involvement with a solitary cyst measuring 2.5 cm in diameter. Gradual shrinkage and radiographic disappearance of the lesion was evident in the follow visit accompanied by spinal recurrence.

The economic impact of hydatid cystic disease is enormous. It imposes a significant burden on individuals, families, and communities, even in developed societies. According to data reported in an Italian study, the estimated average national financial burden is €4 million per year.[29]

Informed consent from the patients was obtained by the authors for the purpose of scientific publication, and appropriate ethical clearance was attained.

Conclusion

Hydatid bone disease is an uncommon presentation of a common disease. It is prevalent in Middle Eastern, African, and Mediterranean countries. Control of affected livestock and dogs breaks the cycle of hydatid cystic disease, decreases the economic impact of the disease, and dramatically reduces inadvertent transmission to humans. Early diagnosis facilitates the prompt and optimal management of cases and guards against complications, which include fractures, deformity, paralysis, and anaphylaxis.

Management plans include a combination of preoperative and postoperative anthelminthic drugs, surgical attempts to completely remove the cyst, and optional intraoperative application of the PAIR technique. Long-term follow-up is mandatory to prevent complications and recurrence, and to monitor residual disabilities.

Conflict of interest

The authors have no conflict of interest to declare.

Ethical approval

The research complies with the rule of clinical ethical committee of the Ministry of health Sudan, and the University of Tabuk.

Consent

A consent for approval has been taken from the patients, and their identities have been concealed.

Authors' contributions

The concept and design: AFAF & AAAS. Data collection: AFAF & AAAS. Data analysis: AFAF & AAAS. Manuscript drafting and critical revision for important intellectual content: AFAF & AAAS. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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