Mycetoma revisited

Incidence of various radiographic signs

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ABSTRACT

الأهداف: تحديد وبيان كمية مختلف العلامات الإشعاعية لعدوى الورم الفطري.

الطريقة: أجريت دراسة إسترجاعية شملت 516 مريضاً تم اختيارهم عشوائيا خلال الفترة مابين ديسمبر 1996م وحتى ديسمبر 2004م، في مركز أبحاث الأورام الفطرية في جامعة الخرطوم – الخرطوم – السودان. تم تأكيد جميع الحالات من خلال الفحص السريري وصور الأشعة المبدئية السابقة للعلاج وتحليل الأنسجة.

النتائج: كانت أكثر العلل شيوعا لدى 516 مريضاً هي عبارة عن تورم الأنسجة الطرية بنسبة وصلت إلى 93%، تصلب العظام بنسبة 56%، تسوس العظم بنسبة 32% والتفاعلات بجانب العظم بنسبة 27%. أفادت التقارير بحدوث تمدد العظم لدى 22%، التواء القشرة الخارجي المنشأ لدى 22% والمباعدة بين العظام لدى 10% من المرضى لأول مرة في هذه الدراسة. كما لوحظ ترقق العظام لدى 19%. فقط 3% من المرضى كانت صورهم الإشعاعية الطبيعية.

خاتمة: تعتبر هذه الدراسة هي أكبر دراسة إشعاعية للورم الفطري. يعد الفحص الدقيق لصورة الأشعة فقط من قبل أطباء الأشعة المتمرسين أمر حيويا لأن أساليب التصوير الأخرى غير متوفرة في المناطق التي ينتشر فيها الورم الفطري.

Objectives: To define and quantitate various radiographic signs of mycetoma infection.

Methods: A retrospective study of 516 randomly selected patients seen between December 1996 and December 2004 at the Mycetoma Research Centre of Khartoum University, Khartoum, Sudan. All cases were confirmed by clinical examination, initial pretreatment radiographs, and histopathology.

Results: The most common abnormalities in these 516 patients were soft tissue swelling (93%), bone sclerosis (56%), bone cavities (32%), and periosteal reaction (27%). The incidence of bone expansion (22%), extrinsic cortical scalloping (22%), and fanning of the rays in 10% were reported. Osteoporosis was seen in 19%. Only 3% of the patients had normal radiographs.

Conclusion: Maximal scrutiny of radiographs alone by experienced radiologists is vital, as other imaging techniques are not available where mycetoma is prevalent.

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Mycetoma is a chronic non-specific granulomatous inflammatory disease. It is a serious health problem in many developing countries.[1-5] Mycetoma is endemic in what is known as the mycetoma belt, which stretches between latitudes 15°S and 30°N.[1-4] However, it was also reported from the developed parts of the world.[6,7] Mycetoma is a peculiar and distinct clinical entity caused by either true fungi "eumycetes," or by actinomycetes gram positive branching aerobic bacteria. Hence, they are named eumycetoma and actinomycetoma, respectively.[1] Painless subcutaneous tumefaction, sinuses formation, and serosanguinous discharge that contain grains are pathognomonic of mycetoma.[1] Many radiological techniques are in use for the diagnosis of mycetoma including conventional radiographs, ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI).[2,8-12] Such scans are too expensive for the patients or the health

authorities in tropical countries. The plain film findings were first described by Davies in 1958.[9] Radiographs were considered most useful.[2] However, MRI was reported to be positive in 90% of the cases compared to 50% positive plain films.[1] Computed tomography was previously considered more helpful than MRI.[2] Ultrasonography is awaiting further experience to be widely accepted.[10] Conventional radiographs have distinct features, which can help in the diagnosis and follow up of patients after treatment.[2,8,9] Recently, a staging classification of pedal mycetoma infection was possible on radiographic examination alone.[13] The aim of this review was to define and quantitate the various radiographic signs of mycetoma infection to improve detection rate, and maximize the benefits of simple radiographs, which may be the sole available diagnostic modality in many endemic areas.

Methods. This retrospective study was conducted at the Mycetoma Research Centre, University of Khartoum, Khartoum, Sudan. It included randomly selected 516 patients with confirmed mycetoma infection seen over a period of 8 years between December 1996 and December 2004. The diagnosis of mycetoma was confirmed by the clinical examination, radiographic appearance of the lesions, and histopathological examination of the surgical biopsies and grain culture. Radiographs were scrutinized for soft tissue swelling, periosteal reaction, and bone lesions. The site, size, and density of the lesions were recorded. The distribution pattern of the lesions was noted. Findings were tabulated for total counts. Only the initial radiographs carried out at the first presentation were included. Reference is made to the recent classification of bone changes in mycetoma.[13] Follow up radiographs after surgery or medical treatments were excluded from the study. Other imaging modalities, which were rarely available, were not included in this study. This review was approved by the Mycetoma Research Centre Committee.

Results. This descriptive study included 516 patients of whom 399 (77%) were males, and 117 (23%) were females. Their ages ranged between 4 and 65 years with a mean of 27.4 years. The most common site of mycetoma infection in this series was the lower limb in 463 (89.7%) of patients, followed by the upper limb in 41 (8%), trunk and pelvis in 7 (1.3%), and skull in 5 (1%). The foot was the most commonly affected single body part in 398 (77%) of the patients. In most of the 516 patients, 502 (97.2%) had one or more radiographic signs of mycetoma. The spectrum of radiographic signs encountered in this study were summarized in **Table 1**. The most common abnormality was the presence of soft tissue swelling, which corresponds to stage O of the new classification (**Figure 1a**). Fanning of the rays due to an expanding granuloma represents stage I (**Figure 1b**). Reactive bone sclerosis was the second most common findings corresponding to stage II (**Figure 2**). It was diffuse in 110 (21%) of the 516 patients, focal in 59 (11%), or surrounding the bone cavities in 113 patients (22%). Actual bone invasion was the third most common finding in this series. The small bones of the feet were involved in 239 (46%) patients. Commonly, multiple bones are affected at the time of presentation in 174 (33.7%) of all the patients. The disease was localized to a single bone in 65 (13%) of the patients corresponding to stage III (**Figure 3a**). It involved 2-5 adjacent bones in 110 patients (21%) (stage VI, **Figure 3b**), 6-10 small bones in 43 patients (8%, stage V) (**Figure 4**), and dramatically involved more than 10 bones of a single foot in 21(4%) of the patients (stage VI, **Figure 5**). In the foot, the most common affected region was the forefoot, which was documented in 185 (36%) of the patients (**Figures 1, 2 & 3b**), while the hind foot was the second affected pedal site in 101 (20%) patients (stage V, **Figure 4**). The midfoot was the third affected site in 91 patients (18%). Extensive subtotal destruction of the foot (stage V) was seen in 14 patients (3%), and total disruption of the whole foot (stage VI) was evident in 7 (1%) of the patients (**Figure 5**). In this study, the majority of bone cavities were multiple. It was single in 28 (5.4%) of the patients (**Figure 3a**), ranging between 2-6 in number in 64 patients (12.4%), between 6 and 19 in 46 patients (9%) (**Figure 4**), 20-39 in 18 patients (3.5%), and 40 in 7 patients (1.3%). Bone cavities were usually larger than 5 mm (**Figure 4**). The actual size of the cavities were as follows: in 41 (8%) patients it measured 1-5 mm, in 56 patients (11%) it measured 5-10 mm, in 58 patients (11%) it measured 11-30 mm, and 8 patients (1.5%) had cavities larger than 30 mm in

Table 1 - Common radiographic signs of mycetoma infection (N=516).

Radiographic signs	n	(%)
Soft tissue swelling	479	(93)
Bone sclerosis	289	(56)
Actual bone invasion	239	(46)
Extrinsic cortical erosion	197	(38)
Bone cavities	163	(32)
Periosteal reaction	135	(26)
Bone expansion	115	(22)
Extrinsic cortical scalloping	115	(22)
Osteoporosis	100	(19)
Joint involvement	99	(19)
Fanning of the rays	54	(10)
Normal appearances	14	(3)

Figure 1 - Radiographs of the foot of **a**) a 25-year-old male showing soft tissue swelling of the big toe due to mycetoma infection (stage 0). No bone involvement. **b**) a 29-year-old male who presented with soft tissue swelling. There is early fanning of the first ray without bone invasion due to expanding granuloma (stage I).

Figure 3 - Radiograph of **a**) a 36-year-old woman showing erosion of a single phalanx (stage III) **b**) the forefoot of a 24-year-old male showing numerous small cavities affecting multiple bones at the same time. Note the speculated periosteal reaction and the toothbrush pattern. The disease has spread along a single ray crossing joints (stage IV).

Figure 2 - Radiograph of a 31-year-old man showing diffuse metatarsal and tarsal bones sclerosis due to irritation and impending invasion (stage II).

Figure 4 - Radiograph of the hind foot of an 11-year-old child showing multiple cavities due to invasion of adjacent bones in the hind foot (stage V).

size. In 82 (16%) patients the periosteal reactions was thick and well organized indicating chronicity, and in 17 patients (3%), it was toothbrush (**Figure 3b**). Sunray pattern was seen in 12 patients (2%), and a lamellar type in 4 (0.8%). Codman's triangle was seen in 8 patients (1.5%). Osteoporosis was diffuse in 48 (9%) of the patients, periarticular in 23 (4.5%), focal in 20 (4%), and massive in 9 patients (2%).

Discussion. Previous reports on radiographic features of mycetoma are few and involved small numbers of patients (**Table 2**).[2,8,9,12-16] A lot of clinical studies were carried out in Sudan, where this review was also conducted,[1,10,17-19] as well as in India,[20] and Senegal.[21] In this retrospective study, only 3% of our patients had normal radiographs. This is a lower negative yield compared to the previous reports of 28-50% normal radiographs in mycetoma patients.[2,8] Other researchers reported 100% incidence of soft tissue swelling on radiographs, however, their sample sizes of 16 and 30 patients[2,15] were very small compared to 516 in this study. Picking up soft tissue swelling (stage 0), which is by far the most common radiographic sign, needs proper techniques with soft tissue exposure, or the use of the simple and forgotten bright spot light for optimum viewing. Where available, CT or MRI might pick subtle changes when the radiograph is normal. In some rural health facilities, radiographs are badly stored, improperly transported, and are prone to have artefacts. However on perseverance, the relevant information can be extracted.

Initial scrutiny or over-reading by experienced radiologists of simple radiographs raise the detection rate of abnormalities. Bone sclerosis observed in this series matches the incidence in other series.[2,8] This corresponds to stage II (**Figure 2**) of the impending bone invasion (**Figure 2**). Bone cavities were relatively of large size and few in number (**Figure 4**). This can be explained by the fact that eumycetoma is predominant

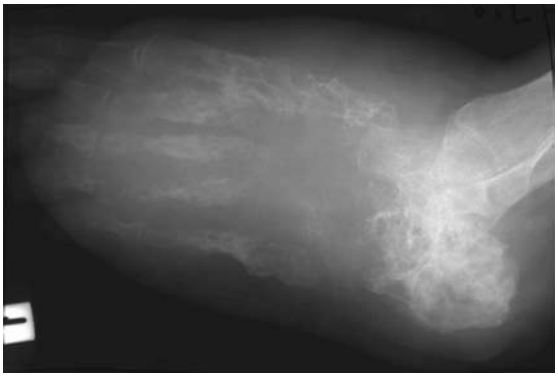

Figure 5 - Radiograph of a 35-year-old male showing total disruption of the foot due to multidirectional spread with multiple skin nodules and invasion of all bones in the foot (stage VI).

Table 2 - Description of radiographic signs of mycetoma.

Studies	N	Year	Country	Reported radiographic signs									
Carrol[14]	1	1949	USA	Decalcification	Atrophy	Rounded bone destruction	Half-moon cortical defects	Fusion	Joint destruction				
Davies[9]	21	1958	Uganda	Loss of definition of cortical margins	Extrinsic erosion	Periosteal reaction	Bone sclerosis Multiple lucencies	Distal osteoporosis	Melting snow	Gap between bones	Soft tissue cavities		
Lewall[2]	30	1985	KSA	Periosteal reaction (67%)	Sclerosis (53%)	Endosteal reaction (50%)	Cortical erosion (43%)	Translucencies (33%)	Joint destruction (27%)	Moth eaten (17%)	Osteoporosis (3%)	Bone lysis (3%)	Soft tissue swelling (100%)	Normal bones (27%)
Bendl[8]	31	1987	KSA	Periosteal abnormalities (72%)	Sclerosis (55%)	Osteolytic defects (34%)	Joint destruction (28%)	Normal bones (28%)					
Shraif[11]	18	1991	KSA	Soft tissue distortion or tumor	Cortical thinning	Spiculated periostitis	Osteopenia						
Czechowski[12]	20	1997	UAE	Lytic destruction	Sclerosis	Remodeling of bone	Normal (50%)						
Al-Ali[15]	16	1997	KSA	Density changes (68%)	Scalloping (31%)	Erosion and destruction (43%)	Periosteal reaction (37%)	Soft tissue changes (100%)	Cystic changes with sclerosis (35%)				
Khatri[16]	70	2002	Yemen	Bone involvement (39%)	Cavities (21%)	Periosteal reaction (34%)	Sclerotic (8%)						

USA - United States of America, KSA - Kingdom of Saudi Arabia, UAE - United Arab Emirates

in our series.[1] Mycetoma and the feet are synonymous. The high incidence of pedal mycetoma in 77%, and the preference of the fore foot in 36% of the affected feet are in line with other reports.[2,16] Incidence of hand affection (6%) (**Figure 3a**), is less than a previous report of 12%.[1,22] This is perhaps due to a change from hand cultivation to semi-mechanized farming.

The majority of patients (73%) had some form of bone involvement at the time of the first presentation (stage III or more). This reflects how sinister is mycetoma by burrowing deeply to reach the bones before the patient becomes aware of the disease. It has been reported that mycetoma is usually painless.[1] Bone expansion was well known, but the incidence in 22% is the first time ever to be reported. Likewise, 22% overall higher incidence of extrinsic cortical scalloping (stage I) was not previously reported.[12] These are important signs indicating impending bone invasion. Surgical intervention at this stage may be more localized, and will avoid more mutilating surgical excision.

The incidence of osteoporosis in this series (19%) is higher than the previous report of 3%.[2] This higher detection rate could be due to improved radiographic technique, and the higher index of clinical suspicion. The incidence of joint involvement (19%) in this series (stage IV or more), is less than that reported previously (28%), and the explanation of this is unclear.[2,8] It may be due to the property of the prevalent eumycetoma fungal agent not to attack the joints in the earlier stages, unlike the bacterial agents of actinomycetoma. Fanning or gap between the metatarsal or metacarpal rays (stage I) in 10% is an early warning sign of granuloma expansion.[9] This is an important marker of the best time for surgical intervention, as the surgery will be limited to the soft tissues, avoiding bone resection or amputation. The incidence of this sign was not previously reported.

Two previous researchers[2,8] reported a much higher incidence of periosteal reaction of 67 and 72% compared to 27% in our series (stage II). Those reports were from the same center in Saudi Arabia where there is a higher incidence of actinomycetoma. Certain types of periosteal reaction can cause some concern when they take the shape of Codmans triangle, lamellar pattern, or sunray speculation, which may be indistinguishable from osteosarcoma or Ewings tumor.[22] These were detected in 4.5 % of all patients.

The limitation of this study was the lack of comparison to other imaging modalities.

In conclusion, maximal scrutiny or over-reading of radiographs by experienced radiologists is vital for diagnosis, staging or classification of the disease, because mycetoma is prevalent where other imaging techniques are not available. Diagnosis at an earlier stage would prevent progression of the disease to a disfiguring or disabling stage. Further studies to assess the significance of each radiographic sign and their impact on the medical or surgical decision-making is necessary.

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