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Letter to Editor

An update and issues on "Aneurysmal bone cyst (ABC)"

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Sir,

At the outset, I would like to congratulate the authors of the article published in your journal in the current issue on - Aggressive lytic lesion of lesser trochanter proximal femur in an eighteen year old male – A case report, which was published in the July'20 issue of this journal. However, certain serious and pertinent concerns are required to be addressed for the knowledge update and benefit of the readers of this article.

It is pertinent to highlight the description given by World Health Organization (WHO) mentioning ABC as "an expanding benign osteolytic lesion having blood-filled spaces of different sizes which are separated by connective tissue septa containing trabeculae or osteoid tissue and osteoclast like giant cells.[1]"

It should be highlighted that although ABC is a separate entity but clinician should consider differential diagnoses like unicameral or solitary bone cyst, chondroblastoma, and fibrous dysplasia with special emphasis on the likelihood of Giant cell tumour or telangiectatic variant of osteosarcoma. Author should have considered these differential diagnoses.

There is no mention of blood investigations by the author as Erythrocyte sedimentation rate (ESR) and Alkaline phosphatase levels may be increased in ABC.

Routine plain radiographs which are done in these cases usually demonstrates a large cystic and eccentric expansile subperiosteal osteolytic lesion with fine septations and an eggshell-appearing bony rim surrounding the lesion which is characteristically defined as "blown out soap bubble appearance". This appearance is characteristic of ABC; author has neither described it nor is the radiograph shown in the article classical of ABC.

Literature classifies these lesions based on Capanna etal classification based on radiographic findings;[2] where Type I is Central metaphyseal lesion with intact or slightly expanded bone; Type II involves the entire segment of bone with expanded bone and thinned out surrounding cortices; Type III lesion is an eccentric metaphyseal lesion with no or minimal expansion of the cortex; Type IV lesion have subperiosteal extension with no or minimal cortical erosion and Type V lesions are metadiaphyseal in location with expanded bone, cortical penetration and extension into cancellous bone. Author neither has mentioned it nor classified the lesion in the case report presented here.

Literature also shows Lodwick radiographic grading with bone destruction,[3–5] which is a well defined method to express the severity on plain radiographs. It is pertinent to mention it here for updating the knowledge of the readers of this article.

Computed tomography (CT) of the lesion helps in confirming the characteristic features which have been described for ABC on plain radiographic evaluation especially internal septations, egg shell appearance of the rim of the lesion and the fluid-fluid levels within the cysts,

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which are caused by the separation of cellular material and serum within the cystic spaces.[6] The diagnosis is seriously questioned in absence of such findings both on radiographs and CT as mentioned by the author. Magnetic resonance imaging (MRI) can further add to the findings on plain radiography and CT, by revealing blood within the multiloculated cystic lesion and its extension into surrounding soft tissues.[7] These MRI findings which are helpful in diagnosing ABC are conspicuously absent in the description mentioned in the case report.

It is worthy to note that the clinical and radiologic features together are sufficient to make a presumptive diagnosis of ABC. However, histological diagnosis is considered essential before planning the definitive management of the lesion. The histology is likened to a "blood-filled sponge". It is composed of blood filled interlaced cystic cavernomatous spaces, the walls of which are composed of fibroblasts, myofibroblasts, osteoclast type of giant cells, osteoid tissue and woven bone. The distinguished histological findings are important to confirm the diagnosis of ABC, which this article conspicuously lacks. It is desirable to mention Enneking classification for benign lesions. The lesion presented here should have been staged (i.e. stage 1, 2 or 3). Enneking has differentiated it into three stages like Latent (inactive), Active and Aggressive, with increasing severity for the benefit of the readers.[2,8]

Thus in combined absence of classical radiological findings on plain radiograph, CT, MRI and histology as mentioned above; author's labelling this case as ABC raises serious doubts.

Readers should note due to the unusually large bony involvement, osteolytic and expansile characteristic of the lesion, intervention is preferred over mere observation. Currently trend is to use sclerotherapy with trials of wide range of sclerotherapy agents. Intralesional injection of drugs is preferred because it offers a least invasive therapeutic option. It works by causing damage to vascular endothelium initiating the cascade of events resulting in healing of the lesion. Various agents have been described for sclerotherapy like percutaneous intralesional administration of ethanol 96%, Aethoxysclerol 3% (polidocanol- hydroxypolyaethoxydodecan),[9] Ethibloc (a hydroalcoholic radioopaque solution of zein),[10] triamcinolone acetonide,[11] liquid absolute alcohol and absolute alcohol ge.[12,13] Intravenous denosumab[14] has been tried for ABCs in anatomically critical sites. Literature shows the use of percutaneous cryoablation & embolization with N-2-butyl-cyanoacrylate under image guidance has been used successfully in patients of spinal ABC.[15]

It is worthwhile to mention the recent modalities in the management of ABC. Studies have shown the use of percutaneous injection of bone marrow derived concentrated stem cells[16] and even whole bone marrow to be effective in aneurismal bone cyst resolution.[17,18] It is presumed that the bone marrow derived stem cells differentiate into cells of osteoblastic lineage leading to healing of ABC lesion.

1. Conflict of Interest

None.

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