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New innovative treatment option of aneurysmal bone cysts with Denosumab

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Aneurysmal bone cysts (ABC) are expansible destructive tumors which are positive for markers of osteoclasts resembling Giant Cell Tumors (GCT). The treatment of ABC implies surgical resection, curettage and cavity filling, embolisation, fibrosing agent injection or radiotherapy. These options can be unsatisfactory in children and adolescents with lesions in critical locations, e.g. in the spine, implying the need for innovative therapies.

Denosumab is a human monoclonal antibody that inhibits osteoclast function by blocking the cytokine receptor activator of NFkB ligand. Satisfying results of Denosumab in treatment of GCT and the immunohistochemical similarities justify the assumption of positive effects on ABCs.

This report is the first description of a therapeutic use of Denosumab in a patient with a spinal ABC. A case of an eight years old boy with a recurrent spinal ABC at C5 after surgery with intralesional tumor resection is described. The interdisciplinary tumor board discussed the remaining treatment options, implying revision surgery, embolisation, radiotherapy, fibrosing agents and an individualized treatment without as yet scientifically proven benefit using Denosumab. Due to relevant disadvantages of surgery (unlikelihood of wide resection, undesirable instrumented fusion in a growing child), radiotherapy (risk of radiation injury) and fibrosing agents (risk of emboli), embolisation was tried, but failed due to an absence of appropriate tumor vascularization. Finally, Denosumab therapy was initiated following extensive information and written consent by the family and approval of the health service.

Denosumab was given at a dose of 70 mg/m² BSA SC q 4 weeks, the dose being adapted from the approved adult dose of 120 mg q 4 weeks. Denosumab therapy was supplemented by appropriate daily oral substitution of calcium and vitamin D.

Since Denosumab therapy started, the patient recovered significantly from pain and neurologic symptoms and is in a healthy condition with no severe side effects. MRI checkup after 2 and 4 months and CT scan after 5 months showed regression of the cystic ABC and a replacement by solid bone marrow-like tissue. A longer follow-up and clinical studies will be needed to evaluate Denosumab for patients with ABC in critical locations.

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