

SKELETAL CYSTIC ANGIOMATOSIS: STABILIZATION DURING BISPHOSPHONATE TREATMENT

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Cystic angiomasia is a rare condition of disseminated multifocal hemangiomatous and/or lymphangiomatous lesions of the skeleton with possible visceral organ involvement. The cause of this condition is unknown. Patients with cystic angiomasia may remain asymptomatic or experience bone pain, even in the absence of pathologic fracture. In the present study we described a 20-year old man, in otherwise excellent health, had a 2-year history of cervical and dorsal back pain. Radiographs of the column showed the presence of multiple cystic lesions in the cervical, dorsal and lumbar vertebral body. A triple-phase whole-body bone scan demonstrated focal increased uptake within, with no other abnormalities. Destruction of bone was confirmed by CT scans and MRI scans showed angiomasia soft tissue formation. A bone biopsy specimen appeared as a simple cyst or multiple communicating cysts. Serum calcium and phosphorous levels PTH and Vitamin D metabolites at the presentation were within the respective ranges. Serum bone alkaline phosphatase was elevated (31.4 U/L; reference range 10-25) and desoxypyridinoline (DPO) in the normal range. For additional studies, we assessed the serum concentrations of the osteoclastic regulators osteoprotegerin (OPG) and osteopontin (OPN) at the baseline and during the follow-up by ELISA (R&D System, USA; Immundiagnostik, Germany). At the baseline OPN and OPG levels were high (OPN: 13±0.5 ng/ml reference value: 5.9±0.6 and OPG 125±0.34 pmol/ml reference value 0-30). Patient started an intravenous bisphosphonate infusion (30 mg pamidronate) every month. A follow up after 6 months therapy showed an improve of mobility and back pain of the patient. Imaging studies including conventional X-ray and CT scans after 3 and 6 months of treatment did not show and progression of bone destruction. After initiation of bisphosphonate treatment serum bone alkaline phosphatase levels normalized. In addition OPN and OPG decreased significantly. In conclusion, an immediate clinical improvement of local pain, an improvement of metabolic resorption and a stable clinical, radiological picture during the first 6 months of follow-up suggests effectiveness of pamidronate treatment alone. Longer-term follow-up and additional studies employing bisphosphonates in skeletal cystic angiomasia will help to evaluate this therapeutic approach in further detail.