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**Idiopathic orbital inflammation following intravenous pamidronate**

Sir, A 71-yr-old gentleman was referred with known Paget’s disease. He had presented previously with chronic intermittent back and left thigh pain. He had been partially deaf for 8 yr and had a transurethral resection of the prostate for benign prostate hyperplasia in 1996. He had been otherwise well. Physical examination showed an enlarged left thigh with reduction in internal rotation of the left hip. There were no other physical signs. The isotope bone scan showed increased activity with a typical Paget’s pattern involving the upper two-thirds of the left femur, right hemipelvis, lumbar spine at L2, thoracic spine at T11 and midecervical spine. Radiology showed Pagetic change in the right hemipelvis and left femur with moderate degenerative disease in both hip joints. Biochemistry showed an elevated serum alkaline phosphatase of 222 IU/l (30–126) but was otherwise normal. Full blood count was normal.

The patient was treated with 90 mg i.v. pamidronate in 500 ml normal saline over 6 h. Treatment was without immediate adverse effects. The following day he developed mild symptoms of an upper respiratory tract infection including a mild fever and nasal serous discharge. Five days following treatment he presented at the ophthalmic casualty department with a 4-day history of watering and swelling of his right eye associated with severe pain. Visual acuity was 6/9 in the right and 6/6 in the left. He had a right-sided proptosis with extraocular movement limitation, conjunctival chemosis and diplopia. He was felt to have orbital cellulitis related to ethmoidal cellulitis and was initially treated with i.v. antibiotics. His colour vision then became severely affected and he could only read the first test plate of the Ishihara chart on the right with 12/16 plates on the left. His visual acuity deteriorated quite severely and was recorded as hand movement only 4 days after his ophthalmic presentation. A computed tomography scan showed enlarged rectus muscles on the right with proptosis and oedema of the soft tissues including eyelid and cheek structures. There was a little mucosal thickening posteriorly in the maxillary antra. Thyroid function tests were normal. A presumptive diagnosis was made of idiopathic orbital inflammation (pseudotumour), antibiotics were stopped and he was treated with systemic steroids. There was a dramatic improvement and within 2 weeks his visual acuity was 6/6 on the right with resolution of his diplopia, and
extraocular movements were now full. The proptosis settled. He was then put on a tapering dose of oral corticosteroids, which were stopped 1 month later. On review 2 months later he was completely well and has remained so since. His back and hip pains settled completely with treatment and his serum alkaline phosphatase reduced to within the normal range. A repeat isotope bone scan showed virtual complete resolution of abnormal activity.

The time course of the ophthalmic illness suggests that this gentleman showed a previously undescribed ophthalmic complication following i.v. pamidronate. A viral or bacterial origin may be possible but was felt unlikely by the supervising ophthalmic surgeon. Occasional other ocular complications have been previously described following pamidronate. These include uveitis [1], episcleritis, scleritis, or conjunctivitis [2, 3]. The mechanism of the ophthalmic complication is unknown but amino bisphosphonates can increase the numbers of certain T-cell subsets and thus affect cytokine release. This has been postulated as a mechanism for the known acute-phase reaction with pamidronate [4].

It is possible to speculate that pamidronate-induced cytokine release in the eye muscles was the mechanism of the described ophthalmic complication. This case enlarges the range of ophthalmic complications of i.v. pamidronate and further illustrates that caution should be exercised regarding i.v. treatment. Whether this is a dose-dependent effect is obviously unknown.

P. J. RYAN, R. SAMPATH
Osteoporosis Unit, Department of Nuclear Medicine, Medway Hospital, Gillingham, Kent ME7 5 NY, UK.
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Correspondence to: P. Ryan.