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Case report

Intravenous sodium thiosulfate for treating tumoral calcinosis associated with systemic disorders: Report of four cases

Arthur Mageau [a], Vincent Guigonis [b], Voa Ratzimbasafy [c], Thomas Bardin [a,d], Pascal Richette [a,d], Pablo Urena [e], Hang-Korng Ea [a,d,*]

[a] Pôle locomoteur, service de rhumatologie, centre Viggo Petersen, hôpital Lariboisière, AP–HP, 2, rue Ambroise-Paré, 75010 Paris, France
[b] Service de pédiatrie, hôpital de la Mère et de l'Enfant, 8, avenue Dominique-Larrey, 87000 Limoges, France
[c] Service de pharmacie, hôpital de la Mère et de l'Enfant, 8, avenue Dominique-Larrey, 87000 Limoges, France
[d] UFR de médecine, université Paris 7 Denis Diderot, Paris Cité Sorbonne, 16, rue Henri-Huchard, 75018 Paris, France
[e] Service de néphrologie et dialyse, Ramsay-Générale de Santé, clinique du Landy, 23, rue de Landy, 93400 Saint-Ouen, France

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ABSTRACT

Intravenous sodium thiosulfate (ivSTS) is a promising new therapeutic option for calciphylaxis related to end-stage renal disease. However, its effect on tumoral calcinosis (TC) complicating autoimmune connective-tissue diseases has been scarcely described. We report here 4 cases (3 adults and 1 child) of TC treated with ivSTS. TC was secondary to CREST syndrome, dermatomyositis (1 adult and 1 child) and systemic erythematous lupus and involved multiple sites in all cases. In all 4 patients, TC was responsible for joint pain, reduced mobility, inflammatory flares and skin fistulations. One patient experienced difficulty sitting due to the pain induced by calcified lesions on the buttock; another patient had major disability, moved only with wheelchair and was under opioid treatment for pain. For all patients, treatment with several medications before STS was unsuccessful. The 3 adults received at least 6 cycles of ivSTS (20 g/d, 5 days/month) and the child received a daily infusion of 17 g STS during 1 month then a 9-g/d infusion during 3 months. Two adults and the child showed clinical improvement with STS treatment and the third adult felt disappointed and stopped STS treatment after 6 months. The child also stopped STS after 6 months due to vomiting. In one patient, an intensive regimen of ivSTS (20 g every 2 days) controlled recurrent flares and fistulations. Unfortunately, TC remained unchanged. Further studies are needed to decipher how STS modulates ectopic calcification, the optimal regimen and posology.
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1. Introduction

Tumoral calcinosis (TC) is a frequent complication of autoimmune connective-tissue diseases including systemic sclerosis, CREST syndrome, dermatomyositis (DM) and systemic lupus erythematosus (SLE) [1]. Its prevalence can reach 17%, 25% and 30% to 70% in SLE, systemic sclerosis and DM, including juvenile DM, respectively [2]. Calcinosis can cause pain and skin ulcerations that greatly affect quality of life and may be complicated by infections. TC management is a real challenge because of no efficient drug available. Several therapeutic options that have been used without consistent efficiency include high-dose calcium-channel inhibitors, bisphosphonate, probenecid, colchicine, rituximab and anti-tumor necrosis factor alpha [2,3].

The efficacy of intravenous sodium thiosulfate (ivSTS) to treat calciphylaxis in patients with end-stage renal disease (ESRD) or on hemodialysis has given a promising therapeutic option for other conditions with ectopic calcifications [4]. Previously, we successfully treated uremic tumoral calcinosis during ESRD with ivSTS [5,6]. We report here 4 cases of connectivitis-related TC treated with ivSTS.

2. Case reports

2.1. Case 1

In 2003, CREST syndrome was diagnosed in a 60-year-old woman with dystrophic calcinosis, Raynaud's phenomenon, sclerodactyly and gastroesophageal reflux. Antinuclear antibodies were positive at 1/640 without specificity. TC lesions were first located

* Corresponding author. Pôle locomoteur, service de rhumatologie, centre Viggo Petersen, hôpital Lariboisière, AP–HP, 2, rue Ambroise-Paré, 75010 Paris, France.
E-mail address: korngea@yahoo.fr (H.-K. Ea).

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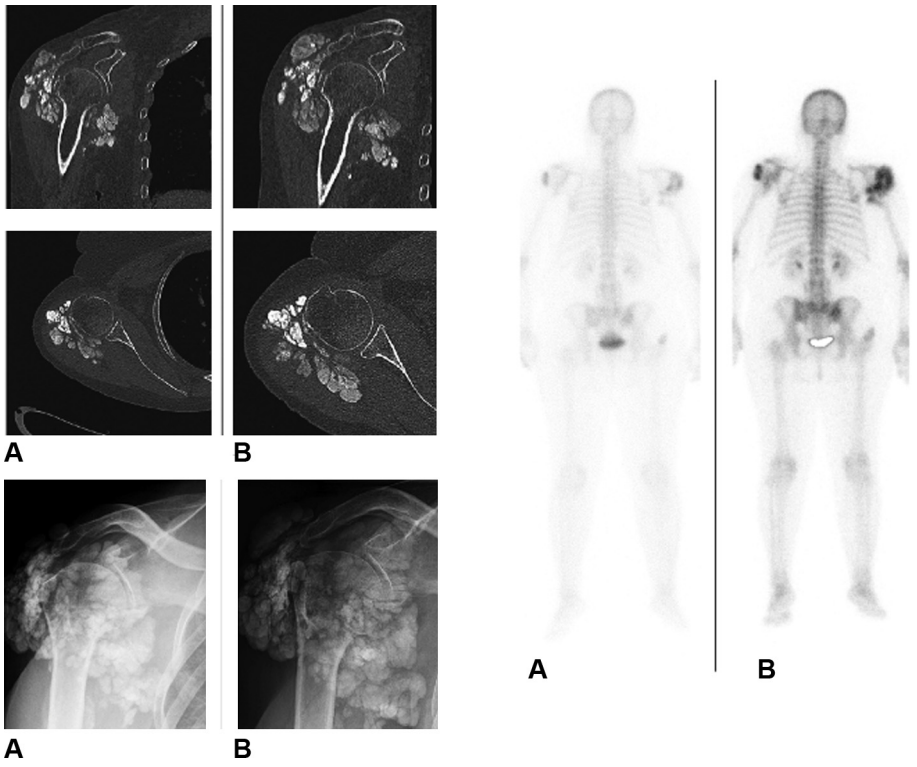

Fig. 1. Shoulder calcinosis during CREST syndrome before (A) and after 6-month treatment with sodium thiosulfate (STS) (B).

on the left hip area and then on both shoulders (Fig. 1). They were responsible for disability, pain, skin ulcerations and joint mobility reduction.

Surgical excision of the hip TC was needed to decrease disability and joint limitation. Several medications were used without effect (probenecid, 2 g/d from 2011 to 2014), diltiazem (300 mg/d from 2003 to 2015) and celecoxib, which decreased the pain, so treatment with ivSTS was proposed. Authorization to deliver this treatment was obtained from the French drug administration authority. The patient received 6 cycles of ivSTS, 20 g/d for 5 days, from February to July 2014. Infusion cycles were given every 4 weeks. Percutaneous STS was also used at the same time, with one application every day. After 6 months of treatment, the patient reported no improvement in pain or joint mobility, and she decided to stop the treatment. Shoulder TC lesions remained unchanged, as evidenced on CT performed before and after 6 cycles of ivSTS (Fig. 1). No adverse event was observed during treatment.

2.2. Case 2

In 2009, TC developed on the right elbow and buttocks of a 40-year-old woman with DM associated with Goujerot-Sjögren syndrome. TC lesions were particularly painful and responsible for major disability and pain when sitting. Several treatments were given without success: colchicine from 2011 to 2012 at the maximum dose of 2 mg/d, which was complicated by frequent diarrhea; 3 different calcium channel blockers (diltiazem 120 mg/d; nicardipine 50 mg/d and nifedipine 30 mg/d); and one bisphosphonate compound, pamidronate 60 mg/month for 6 months, from August 2011 to February 2012. The patient agreed to ivSTS, which was authorized by the French drug administration. A 5-day infusion/month of 20 g/d STS was given from October 2012 to February 2013. Between each cycle, the patient received topical STS daily and oral STS 1.5 g (pills of 500 mg, prepared by the hospital pharmacy).

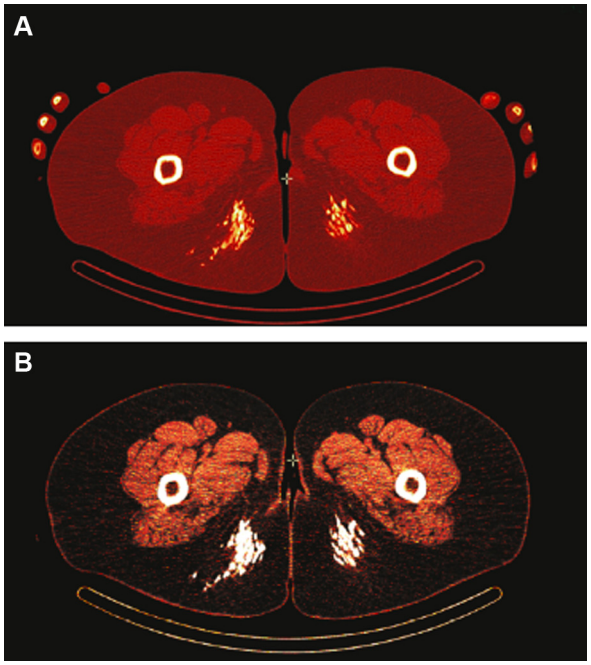

Fig. 2. Buttock calcinosis during dermatomyositis remained unchanged after STS treatment.

After 6 oral and ivSTS cycles, no clinical improvement was observed. In contrast, bone scintigraphy showed increased activity of the TC on buttocks (Fig. 2). Oral and ivSTS were stopped, and other drugs were tried without efficiency: 5 mg intravenous zolendronate/month for 6 months, topical STS, probenecid (2 g/d) and anakinra (recombinant interleukin-1 receptor antagonist), because the patient experienced frequent inflammatory flares and skin ulcerations. Because the TC progressed, it became more

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