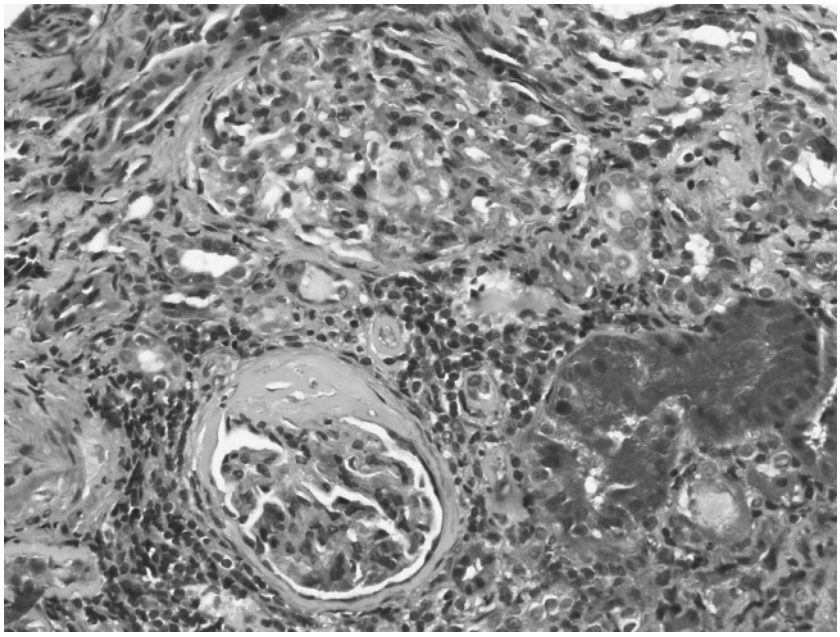

Fig. 1. Optical microscopy. Hematoxilin-eosine. 100×: chronic tubulo-interstitial damage with some inflammation, and segmental sclerotic lesion in one glomerulus.

is apparently rare and mainly seen in patients with mild pre-existing renal impairment [3,4]. In the same manner as other reverse transcriptase inhibitors, RBV could compete with tenofovir for the human organic anion transporter-1 (hOAT1), on the basolateral membrane of the proximal tubules, resulting in an increased tenofovir concentration and tubular toxicity [1,5].

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Advance Access publication 13 May 2008

A new case of Finnish-type congenital nephrotic syndrome, neuromuscular symptoms and early death

Sir,

The children affected by Finnish-type congenital nephrotic syndrome (FCNS) frequently show muscular hypotonia and neurological development retardation, which usually ameliorate after kidney transplantation [1,2,3,4]. However, Laakkonen H *et al*. recently described six FCNS patients with a severe dyskinetic cerebral palsy-like syndrome with dystonic features, athetosis and a hearing defect. Four of the patients died between 1 and 4 years of age in different circumstances; the others survived with severe neurological deficits. In spite of normal metabolic investigations, Laakkonen *et al*. hypothesize that the complex neurological picture was due to mitochondrial dysfunction [5].

We observed a child with FCNS and similar dyskinetic symptoms who died suddenly when he was 4.5 years. He was born at 34 weeks; elevated α-feto-protein was detected in maternal serum (1100 ng/ml) and amniotic fluid; the placenta was 1200 g with a placenta weight/newborn weight ratio of 0.44. Neonatal hyperbilirubinaemia, max 5.5 mg/dl (5.3 mg/dl non-conjugated and 0.2 mg/dl conjugated), was observed. At the age of 3 weeks, secondary hypothyroidism (T4 0.63 ng/dl and TSH 13.1 μU/ml; nv: T4 0.8–1.9 ng/dl, TSH 0.4–4 μU/ml) was diagnosed; thyroxine 37.5 μg/day normalized thyroid hormones (T4 1.2 ng/dl and TSH 3.4 μU/ml). Laboratory parameters also showed heavy proteinuria (>300 mg/Kg/day) and severe hypoalbuminaemia (0.78 g/dl). The clinical diagnosis of FCNS was later confirmed by molecular study for NPHS1 locus. At the age of 3

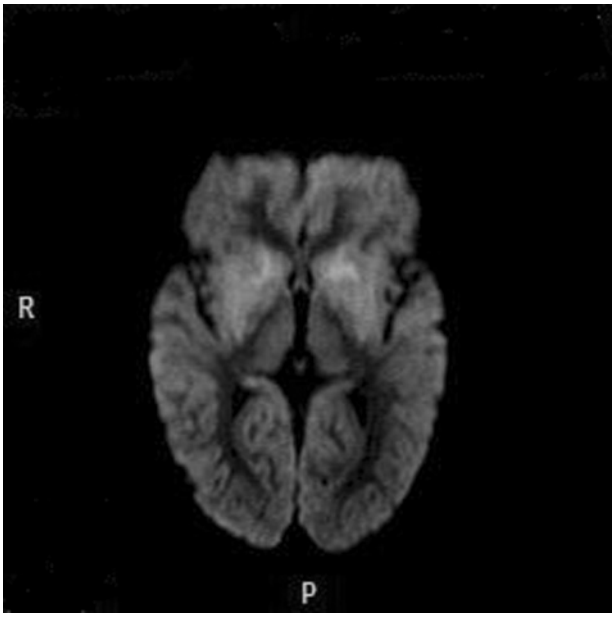

Fig. 1. MR imaging in the first 24 h after acute ipoxic–ischaemic brain damage. Axial apparent diffusion coefficient map shows a reduced ADC value (0.41) in posterior putamen and globus pallidus, and a high ADC value in anterior putamen and head of caudate nuclei (0.73), indicating the prevalence of T2 effect. The relatively high ADC value indicates chronic impairment of anterior portion of basal ganglia.

the patient developed ESRF and began peritoneal dialysis. When he was 4 years, in spite of a satisfactory cognitive and emotional development, the child showed severe neuromuscular deficit with focal hand dystonia and athetosis, inability to keep his head and trunk upright, or walk. He had no hearing defects. No metabolic investigations were performed. At the age of 4.4, after 6 months of treatment with growth hormone, the child weighed 14 kg (10°C) and was 96 cm tall (10°C). During a febrile illness, he manifested apnoea, followed by cardiac arrest, prolonged brain ischaemia and irreversible coma. An MRI performed in the first 24 h showed, besides brain atrophy, T2 hyperintensity on basal ganglia. Diffusion-weighted images showed an extensive signal alteration indicating water diffusion restriction in the lenticulostriate nuclei and the caudate nuclei head with low ADC values (4.1 ± 0.15 SD) in the posterior portion of the lenticulostriate nuclei. The anterior portions of lenticulostriate nuclei showed higher ADC values (7.3 ± 0.43 SD) in the same regions of hyperintensity on T2 weighted images (Figure 1). Therefore the hyperintensity might be related to abnormalities of metabolic mechanisms existing before the hypoxic injury [6]. The child died after 2 months of coma.

Like the cases described by Laakkonen H *et al*., we can exclude prenatal or neonatal complications causing the neurological impairment in our patient. However, our case showed several different features, such as the more severe muscular hypotonia, adequate growth, growth hormone therapy and normal hearing. In spite of these differences, the MRI findings and the unexplained sudden event causing early death strongly suggest that our patient may have been suffering from the new clinical entity that Laakkonen *et al*. call NPHS1 with muscular dystonia and athetosis.

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Viruses may trigger allopurinol hypersensitivity syndrome

Sir,

A relationship between allopurinol hypersensitivity syndrome (AHS) and viral infections, especially with human herpes virus (HHV)-6, has been suspected since the first report of AHS in 1970 [1]. Although participation of other opportunistic viral infections has also been reported, the mechanisms by which any of these contribute to AHS are still unknown. We report a case of a 39-year-old woman receiving peritoneal dialysis due to lupus nephritis who developed severe AHS, which may have resulted from influenza virus infection. She experienced a systemic skin rash with high fever just after administration of allopurinol. Both hypereosinophilia and high titre influenza virus type A-specific antibody were present before admission and at hospital day 6. Because antibody titres for HHV-6