Anterior spinal artery syndrome (ASAS) often leads to complete motor paralysis with poor clinical outcome. There is a lack of controlled clinical trials on acute treatment strategies in ASAS. However, systemic thrombolysis with recombinant tissue-plasminogen activator (rt-PA) might be a useful therapeutic option in ASAS. We report the management of a patient with ASAS below thoracic level 10, who was treated with intravenous thrombolysis. An 81 year old patient presented with flaccid paraplegia. After exclusion of aortal dissection, spinal tumour or haemorrhage, the patient was treated with intravenous rt-PA 3 h 40 min after symptom onset. The follow up magnetic resonance imaging (MRI) showed spinal infarction below thoracic segment 10. In the clinical course, the patient partially recovered lower limb muscle strength and was able to walk with assistance. To the best of our knowledge, this is the first case in the literature of ASAS with MRI-proven spinal ischemia and the application of rt-PA. Systemic thrombolysis seems to be justifiable in patients with ASAS after the rule-out of aortal dissection and spinal bleeding.