Clarifications to ''First described case of coma triggered by retrograde venous air embolism: an exceptional but potentially life-threatening situation''☆

Aclaraciones a «Primer caso descrito de coma desencadenado por embolismo aéreo venoso retrógrado: una situación excepcional pero potencialmente letal»

Dear Editor:

We consider it extremely important to make some clarifications regarding our study published in June 2016.[1] The study reported the case of a woman who attended our department in 2013 due to acute cor pulmonale and coma, which were finally attributed to a retrograde venous air embolism through a peripheral intravenous line (PIV). The mechanism of the embolism was not fully understood but we decided to report it due to its singularity and relevance. Our study said: ''Our patient was a 79-year-old woman with no relevant medical history who was admitted due to lower limb cellulitis. While hospitalised, she removed her PIV accidentally when she was getting out of bed; as a result, she experienced a sudden drop in arterial blood pressure, tachypnoea, and a decrease in the level of consciousness.'' So expressed, it may be inferred that there was a spontaneous ingress of air into the venous system coinciding with the loss of the PIV. However, detailed analysis of the case and the subsequent events clearly contradicts this idea; the case is currently under investigation as the pulmonary air embolism was allegedly caused intentionally by a nursing assistant at our hospital, who is currently remanded in custody (the case is being treated as attempted murder: the patient survived for reasons outside the intent of the suspect).[2]

Having reviewed the case, we consider it very unlikely that the ''spontaneous ingress'' of air through a PIV would cause an air embolism of such magnitude in the patient reported due to a number of reasons, including the following: a) for air to enter the PIV, it must be open; however, peripheral lines or catheters by definition should be used in closed systems (to avoid contamination/infection)[3–5]; b) if a PIV is open or removed from the venous access, blood leaks from the vein in sufficient quantities that is easily detected by the patient, family members, and/or healthcare staff, whereas no blood was observed around the line when our patient was attended; c) Valsalva manoeuvres infrequently induce such considerable air ingress through a PIV; as specified in our article, significant air flows ($\geq$ 0.2 L/min) are needed to cause an air embolism[6]; and d) the hypothetical spontaneous air ingress through a PIV does not usually cause subcutaneous emphysema. Our patient presented significant subcutaneous emphysema extending to the root of the upper limb where the PIV was inserted. Emphysema was probably caused by a high-pressure injection of air or gas, which was extravasated out of the venous layer and penetrated the subcutaneous cellular tissue (this probably saved the patient's life).[7–9]

Therefore, it is likely that the ingress of air or gas into the patient's venous system was forced and not spontaneous, and that the forced introduction of air caused the removal of the PIV, rather than the contrary sequence of events.

In short, spontaneous passage of air or gas into the venous system through a PIV or the removal of it does not usually cause a severe systemic air embolism; in such cases, we should consider the possibility that the entry of air may have been forced.

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Subacute motor neuronopathy associated with Hodgkin lymphoma☆

Neuronopatía motora subaguda asociada a linfoma de Hodgkin

Dear Editor:

Paraneoplastic neurological syndromes (PNS) occur in approximately 1% of patients with solid tumours, and are thought to be rarer in haematological cancers.[1] The PNS most commonly associated with Hodgkin lymphoma (HL) is cerebellar degeneration, whereas peripheral nervous system involvement is more infrequent.[2] Motor neuronopathy (MN) is a rarely reported symptom in patients with HL. We present a case of this association, as well as a brief literature review.

The patient was an 82-year-old man with no relevant medical history who was diagnosed with classical HL after biopsy of cervical adenopathies. He presented no B symptoms. A tumour staging study returned results compatible with stage Ia. He started treatment with doxorubicin, bleomycin, vinblastine, and dacarbazine (ABVD) at 2 months, with neutropaenia and skin lesions appearing as adverse effects. Remission was achieved after 2 months and 2 full cycles of treatment.

The patient was transferred from the emergency department due to rapidly progressive paraparesis, which within 8 days prevented him from walking independently. The patient reported no pain, sensory symptoms, or sphincter dysfunction. The neurological examination revealed predominantly left-sided, distal, asymmetrical paraparesis, with patellar tendon and Achilles tendon areflexia. Stretch reflexes were normal in the upper limbs and plantar reflexes were flexor. Sensory examination showed normal results.

Serology (herpesvirus family, HIV, Lyme disease), immunology (antinuclear antibodies, anti-neutrophil cytoplasmic antibodies, antiganglioside antibodies), and laboratory analysis (including total protein test) yielded normal results. Results from brain and spinal cord MRI scans were normal. A lumbar puncture revealed normal cerebrospinal fluid (CSF) (glucose, 63 mg/dL; proteins, 40 mg/dL; 0 leukocytes/mm^3; no oligoclonal bands). No onconeuronal antibodies targeting synaptic antigens were detected in the serum or the CSF. CSF cytology results were normal. A nerve conduction study (Table 1) performed 2 days after admission revealed severely decreased motor potential amplitudes in the lower limbs, with normal distal latencies. No conduction block was detected. Findings from the sensory conduction study were normal. An electromyography recorded increased insertion activity, with many diffuse fibrillations and fasciculations in the quadriceps, tibialis anterior, and gastrocnemius muscles. Motor action potentials presented markedly increased amplitudes, mild increases in the percentage of polyphasia, and prolonged duration. Recruitment was decreased. Based on these results, we diagnosed severe axonal motor neuropathy with significant signs of denervation and reinnervation. The presence of these 2 signs suggests more prolonged progression than that reported by the patient, and justifies using the term subacute neuronopathy.

The patient's condition improved spontaneously during hospitalisation. He continued on ABVD treatment with no radiotherapy. At one month, he showed a slight neurological deterioration, and a therapeutic trial was conducted with intravenous immunoglobulins, which obtained no response. He remained neurologically stable for the rest of the follow-up period. After 4 cycles and a half of ABVD, the patient presented pancytopaenia with severe neutropaenia, septic shock, and bilateral pulmonary thromboembolism, leading to his death.

MN associated with HL is an infrequent entity and is not included in the European registry of PNS in patients with lymphoma.[2] Most of the reported data are from isolated case reports or small series (Table 2).[3–6] It generally manifests after diagnosis of HL, or even during remission.[3] In general, it predominantly affects proximal or distal areas of the lower limbs. It tends to be mild and to stabilise or even improve without specific treatment,[3] although a favourable response to intravenous immunoglobulins has recently been reported.[4] CSF findings vary, and MRI scans reveal hyperintensities in the anterior horns and contrast uptake in the anterior roots.[4] Anatomical pathology studies show degeneration in the anterior horn of the spinal cord, with variable presence of inflammatory elements.[3,5,6] All these characteristics suggest that the main lesion affects the cell bodies of motor neurons and not the peripheral nerves, hence the term neuronopathy. No associated antibodies have been described.

The aetiopathogenesis of MN associated with HL remains unknown. Exposure to radiotherapy in some of the published cases[3] means we cannot rule out a toxic effect of this treatment. Regarding the possible causal role of ABVD, only vinblastine has been associated with small-fibre sensory neuropathy, and never with motor neuropaties.[7] It is also likely that patients with this diagnosis actually present

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