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Sporadic late-onset nemaline myopathy (SLONM) as a possible manifestation of autoimmune/inflammatory syndrome induced by adjuvants (ASIA)

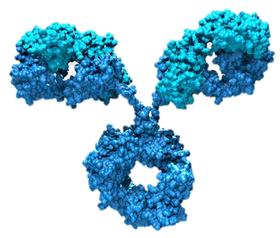
Perna R.[1], *Cevini F.*[1], *Petraroli A.*[2], *Galdiero M.R.* [1,2,3,4], *de Paulis A.*[1,2,3,4]

[1]Specialization School of Allergology and Clinical Immunology, University of Naples Federico II, Naples, Italy

[2]Department of Internal Medicine and Clinical Immunology, University Hospital of Naples Federico II, Naples, Italy

[3]Department of Translational Medical Sciences, University of Naples Federico II, Naples, Italy

[4]Center for Basic and Clinical Immunology Research (CISI), WAO Center of Excellence, University of Naples Federico II, Naples, Italy



INTRODUCTION

- Sporadic late-onset nemaline myopathy (SLONM) is a rare adult-onset, acquired, muscle disease that can be associated with monoclonal gammopathy or HIV infection. The pathological hallmark is the accumulation of nemaline rods in muscle fibers.
- Autoimmune/inflammatory syndrome induced by adjuvants (ASIA) is a syndrome firstly described in 2011 to encompass several autoimmune and inflammatory conditions triggered by various adjuvants, e.g. silicone breast implants.

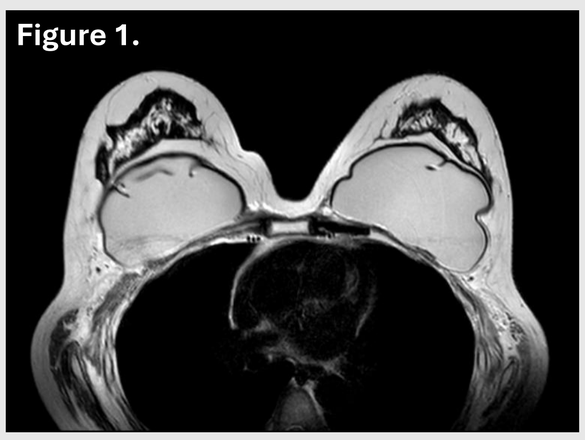
METHODS

We report the case of a 53-year-old Venezuelan woman who was admitted to our clinic presenting with a two-month-history of progressive musculoskeletal pain, extreme fatigue, bilateral proximal and distal muscle weakness and dysphagia.
She presented with no comorbidities. In 2007 she had silicone breast implants for cosmetic purposes.
Physical examination revealed no cutaneous lesions but a widespread deficit of strength in upper and lower limbs. Proximal osteo-tendineous reflexes were weak. No sensitivity deficits and no other abnormalities were observed.

Table 1.		Normal range
C-reactive protein (CRP)	15.1 mg/l	0-5
Creatin kinase (CK)	3057 U/l	0-170
Lactate dehydrogenase (LDH)	588 U/l	135-214 (women)
Aspartate transaminase (AST)	276 U/l	0-32
Alanine transaminase (ALT)	191 U/l	0-33
Anti-nuclear antibodies (ANA)	1:640 speckled pattern	<80

Increased levels of C-reactive protein, creatin kinase, lactate dehydrogenase, transaminases, and anti-nuclear antibodies were found (Table 1).
Extractable nuclear antigen test and anti-double stranded DNA antibodies were negative. All other laboratory tests, including serum protein electrophoresis, were normal. HIV infection and additional microbial infections were excluded.
ECG and echocardiogram were unremarkable. A paraneoplastic condition was ruled out based on tumor markers and whole-body imaging. Swallowing tests, esophagogastroduodenoscopy and esophageal manometry confirmed absent esophageal contractility.

Figure 1.



Despite a negative physical examination, breast ultrasound and MRI (Figure 1) showed a right intracapsular prosthetic rupture.

Electromyography showed signs of muscle damage. A skeletal muscle biopsy revealed myopathic changes compatible with SLONM: cytoplasmic rods were observed in some fibers, along with hypotrophy, occasional myonecroses and myofibrillar disruption.

The patient was treated with Methylprednisolone (40 mg IV twice daily) and IV immunoglobulins (1 g/kg over 3 days). Additional supportive care included total parenteral nutrition and bedside physiotherapy. With steroid tapering, Methotrexate was introduced (15 mg/weekly subcutaneously). Steroid was suspended 5 months after admission.
The patient was recommended prostheses removal.

RESULTS and CONCLUSIONS

- Clinical and serological improvement allowed a slow steroid tapering. CK levels normalised within 6 weeks, and assisted ambulation resumed a month later. Dysphagia resolved slowly: swallowing tests at 4 months post-admission confirmed an improvement and the patient began a semi-liquid diet.
- Our case suggests to evaluate adjuvants exposure in the differential diagnosis of myositis. Noteworthy, SLONM is underdiagnosed and delayed diagnosis and treatment can lead to respiratory muscle involvement, the main cause of death in these patients.
- We therefore suggest that, in patients with silicone breast implants presenting inflammatory myopathy, muscle biopsy and prostheses assessment are warmly indicated.