Received: 17 October 2023 | Accepted: 18 December 2023

DOI: 10.1002/ccr3.8436

Clinical Case Reports Open Access WILEY

LETTER TO THE EDITOR

Sulfasalazine, but not ankylosing spondylitis may be responsible for axonal neuropathy

We read with interest the article by Khalaji et al. about a 45-year-old male with ankylosing spondylitis (AS) first diagnosed 10 years before presentation and treated with sulfasalazine, who developed sensory disturbances (tingling, numbness) for 1 year before presentation in the upper limbs which then spread to the lower limbs.[1] Extensive investigations revealed a sensorimotor, axonal polyneuropathy, but did not provide any information about possible causes of the neuropathy, which is why polyneuropathy was ultimately attributed to AS.[1] The study is impressive, but some points should be discussed.

We disagree with the conclusion that the axonal neuropathy in the index patient was due to AS.[1] Several arguments can be made against a causal relationship between AS and axonal polyneuropathy. First, AS has rarely been described as a cause of polyneuropathy.[2] Second, most patients previously described with AS-associated polyneuropathy were taking sulfasalazine or other neurotoxic medications that may have caused polyneuropathy.[3] Third, the index patient was probably taking sulfasalazine for years, which is known to cause axonal polyneuropathy.[4] Neuropathy has also been reported in patients taking sulfasalazine for ulcerative colitis.[5] Therefore, one should discontinue sulfasalazine and see if the polyneuropathy improves before claiming that AS was the cause.

A limitation of the study in this context is that it is not mentioned how long sulfasalazine was administered. The risk of developing sulfasalazine-induced polyneuropathy most likely depends not only on the dosage but also on the duration of sulfasalazine use.

There is also no mention of how the sensory disturbances were distributed. We should know whether sensory disturbances occurred only distally or also proximally and in a strip-like, glove-like, stocking-like, or area-like distribution.

Another limitation is that long-term follow-up was not reported. We should know how the symptoms developed as the disease progressed, whether they recurred, and what treatment was used.

In summary, it is recommended that sulfasalazine be discontinued in patients with AS and polyneuropathy before attributing the polyneuropathy to AS. If the polyneuropathy does not improve, AS can be blamed after all possible other causes of polyneuropathy have been ruled out.

AUTHOR CONTRIBUTIONS

Josef Finsterer: Investigation; resources; validation; writing – original draft.

FUNDING INFORMATION

None received.

CONFLICT OF INTEREST STATEMENT

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

DATA AVAILABILITY STATEMENT

All data are available from the corresponding author.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

Josef Finsterer

Neurology Department, Neurology & Neurophysiology Center, Vienna, Austria

Correspondence
Josef Finsterer, Neurology Department, Neurology & Neurophysiology Center, Postfach 20, 1180 Vienna, Austria.
Email: fifigs1@yahoo.de

The authors of [Axonal Sensory-Motor Polyneuropathy in Ankylosing Spondylitis: A Case Report] offered no comments.

ORCID

Josef Finsterer https://orcid.org/0000-0003-2839-7305

REFERENCES

1. Khalaji A, Kolahi S, Ghadakchi L, Jafarpour M. Axonal sensorymotor polyneuropathy in ankylosing spondylitis: a case report. *Clin Case Rep*. 2023;11(10):e8038. doi:10.1002/ccr3.8038
2. Knockaert DC, Malysse IG, Peetermans WE. Ankylosing spondylitis. An unusual manifestation of familial Mediterranean fever. Report of a case complicated by amyloidosis and polyneuropathy. *Clin Rheumatol*. 1989;8(3):408-412. doi:10.1007/BF02030357
3. Tekatas A, Pamuk ON. Increased frequency of restless leg syndrome in patients with ankylosing spondylitis. *Int J Rheum Dis*. 2015;18(1):58-62. doi:10.1111/1756-185X.12323
4. Liedorp M, Voskuyl AE, Van Oosten BW. Axonal neuropathy with prolonged sulphasalazine use. *Clin Exp Rheumatol*. 2008;26(4):671-672.
5. Blin O, Sangla I, Jouglard J, Cottin C, Pellissier JF, Serratrice G. Neuropathie axonale et salazosulphapyridine: phénotype acétyleur lent [Axonal neuropathy and salazosulfapyridine: slow-acetylator phenotype]. *Rev Neurol (Paris)*. 1992;148(2):154-156.