

Isolated Toe Tremor with APS: Why Publish?

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My niche discipline, movement disorders, a rather narrow piece of the neurological disorders' spectrum, occupies a minority position in clinical medicine. As our techniques for objective testing for medical disorders increase, continually narrowing the need for clinical expertise, movement disorders, in contrast, remains rooted in the great history of classical neurology, more closely based on clinical acumen, experience and history than on objective diagnostic testing. It is also distinguished by the usual ability of the practitioner to see the effects of therapy. Unlike the common disorders, headache, back pain, epileptic seizures, stroke, concussion, we generally get to see the response to our interventions, and not just the indirect effects, such as better quality of life due to less pain, fewer seizures, etc. We can see better walking, better tremor control, fewer involuntary movements. And, because the movements are always visible, although sometimes episodic and not present in the office, we usually see the responses, or we try to get video recordings of the episodic disorders. We like to share the videos we find interesting. That's one reason for our Video in Medicine report in this issue of RIMJ, "Isolated Toe Tremor Associated with Antiphospholipid Syndrome."

Reporting "interesting cases" is attractive to those of us who try to publish because we think of them as being educational for the reader, interesting to read ("Who would imagine you could see this problem in disease X?"), interesting to

watch, and easy to write. For faculty, the case report can be an important first step in encouraging and mentoring a student, resident or fellow for an academic career. For readers, it may help them diagnose a puzzling disorder. In many disciplines, photos and video images have become commonplace and allow the reader to see a procedure or a treatment effect that they have only read about. They may see a movement they are unfamiliar with and know that the video has been vetted by experts, and not simply something posted on YouTube with unknown provenance.

The problem with trying to publish case reports is that it is often difficult to prove that the problem you're addressing is due to the explanation you propose, since all case reports describe rare things. In the case of our toe tremor case, we noted that tremors of only two toes on one foot had not, to the best of our knowledge, been reported. We could be wrong about this, search engines being not completely reliable. Tremors of the four smaller or all five toes is rare in my experience, but might be seen in Parkinson's disease, whereas tardive dyskinesia or dystonia, often affects the great toe, the four smaller toes or all five. Whenever we see a movement disorder that is rare or unexpected, we try to mimic the disorder to determine if it may possibly be functional, that is, occurring on a "voluntary" or "psychogenic" basis. I saw a patient with an impressive, reportedly involuntary contraction of his soft palate which I thought was functional, based on several

factors, but I reasoned that if I could not mimic it, it might be organic, whereas if it was a psychogenic disorder then I should be able to do it myself. After practicing for a while, I was able to mimic it quite well, supporting my diagnosis, which led to a successful treatment. When an author can't perform the movement, it provides support for the movement being physiologic, hence a movement that is less likely occurring on a functional, that is, psychogenic basis.

The real problem with case reports is connecting the disorder with the proposed etiology. In the toe tremor case, we associated the tremor with an uncommon syndrome, anti-phospholipid antibody syndrome (APS). What if there had never been any movement disorder associated with this autoimmune coagulation disorder? We would likely think to ourselves, like detectives in novels, "I don't believe in coincidences," and, not having an alternative explanation for the tremors, ascribe the movement to the APS. We would not likely have found too many people willing to believe this as an explanation. I see many patients with movement disorders for which I cannot find an explanation. I don't blame their diabetes or their hypertension or their medications. I simply report that I can't provide a diagnosis. When a rare medical condition is followed by a rarer movement disorder we start to assume a connection, but, as we all know, "assume makes an ass of you and me." However, in our case, we argued that APS has been

associated with a variety of movement disorders, fortifying our argument.

What value does the report have? A doctor confronting an unusual disorder for which no cause can be found might come across the report on a computer search and might then check if the patient might have had problems, like miscarriages, or thromboses, to suggest the APS. Several years ago, I saw a patient with unexplained ataxia. He had been evaluated elsewhere with no explanation found.

When I saw him, it was summertime and he was wearing shorts, which revealed his livedo reticularis. I checked his coagulation parameters, which were remarkably abnormal, but before he got to his appointment with a hematologist, he suffered a bowel infarct, requiring surgery.

With sufficient case reports, someone collects enough cases to deduce patterns that might help guide recognition and treatment. In the meanwhile, I invite you to try and wiggle your smallest two toes. ❖

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