



Editorial

Critical Review Ahead of Publication

In a recent commentary in the online website of the Support Group of SMA, it was claimed that scientists had been able to replicate all the Types of human SMA, including Type 4, in a drosophila model of SMA [1]. This sounded bizarre as the different “Types” of SMA are based on the achievement of various motor milestones, such as ability to sit unsupported (“Type 2”) and the ability to stand and walk (“Type 3”). There is no “Type 4” but it was an effort of the clinicians to separate the classical ambulant cases with onset in adolescence, as described by Kugelberg and Welander in 1956 [2], from the later onset “adult” cases with onset after the age of 21. So one can imagine identical twins born a few minutes apart with comparable clinical weakness and genetic mutations but the one being aware of some difficulty before the 21st birthday and the other after.

On reading the manuscript it was obvious that the authors had no idea of the different “types” of human SMA and defined their “Type 4” as “appear healthy and normal at the time of birth with onset of impaired motor function occurring months or years after birth”. It further added “Creating models that mimic this aspect of SMA has been difficult in the mouse, as most genetic manipulations....cause animals to be small, underdeveloped and severely affected at the time of birth.”

So it was quite worrying how this could possibly pass any peer review for publication. The article was published in bioRxiv, a preprint platform provided by the prestigious Cold Spring Harbor Press. bioRxiv is modelled on arXiv, the preprint server set up in 1991 for physicists to upload new ideas for discussion pre-publication.

I have no problem with some bright physicist uploading a brainwave formula such as $E=mc^2$ for comment, but it is a much more worrying situation for basic scientists and biologists, cosseted in an isolated laboratory and with no concept of the human disease, to make spurious claims about replication in animal models and potential benefits from various therapeutic endeavours, often accompanied by hyped up reports in the lay press or the main news media.

BioRxiv has proved very successful for biomedical researchers and has led to the decision to provide a comparable prepublication facility for clinical papers (medRxiv). This has raised alarm bells amongst physicians and in an editorial in JAMA, the editor-in-chief has expressed his concern: “Sacrificing adequate and thoughtful peer review and editorial assessment is a mistake for research in medicine. Timely assessment and dissemination of medical research findings is certainly important, but for most articles, rushing to publication in days or weeks will not improve health outcomes [3].”

It is difficult enough coping with the publicity surrounding current uncontrolled therapeutic efforts in the neuromuscular field. It would be disastrous if there were a *carte blanche* to upload any unregulated claims of therapeutic benefit in animal models or in patients, without peer review. I think we would be providing a useful service to the neuromuscular community if our colleagues could draw attention of our readership to any such uncontrolled studies and bizarre claims, for which I am launching this new forum for **Critical Review Ahead of Publication**.

Victor Dubowitz
Editor-in-Chief

References

- [1] Comprehensive modelling of spinal muscular atrophy in *Drosophila melanogaster*. BioRxiv (a preprint online platform)
- [2] Kugelberg E, Welander L. Heredofamilial juvenile muscular atrophy simulating muscular dystrophy. *Arch Neurol Psychiatry (Chic)* 1956;75:500–9.
- [3] Bauchner H. The rush to publication; An editorial and scientific mistake. *JAMA* 2017;318:1109–10.