

## Decision making about healthcare-related tests and diagnostic test strategies: what “The death of Ivan Ilyich” teaches us



Dear editor,

In the last few years, I have been interested in the field of diagnostic imaging evaluation and, like others in this field, I have been influenced by the 6-step model proposed by Fryback and Thornbury [1]. This model has been a cornerstone in this area, and many papers have been published discussing how to apply it. As an example of the influence of this model, last year, the Journal of Clinical Epidemiology published a series of papers on diagnostic test evaluations [2–6] and the 6-step model was one of the bases for the analyses performed. Also, in a more recent article published this year in the Journal of Clinical Epidemiology, this model was again one of the bases of the analyses [7]. However, although the Fryback and Thornbury model is fundamental for the comprehension of the whole process of diagnostic test evaluation, it has, in my opinion, a philosophical bias in its analytical structure. This bias occurs from step 4 ahead when it considers a diagnostic test useful only if it changes the therapeutic process of the patient and if this change leads to an increment in the amount or quality of life at an acceptable cost. While these assumptions are valid in some situations, as in the evaluation of screening programs, or when the analyses are performed under the perspective of the payers, from the patients’ and physicians’ perspectives in a diagnostic or prognostic evaluation, to use a diagnostic test only when it modifies the therapeutic strategies and the outcome of the patients can be an object of deserved criticism. A classical example is the genetic testing for Huntington’s chorea. Although the use of this test for general population screening is hardly justifiable due to the rarity of the disease and the absence of effective therapies, its use in patients with clinical symptoms suggestive of the disease could be important to confirm or exclude the diagnosis [8,9]. Here, it is important to say the obvious: to know that there is nothing to do for a medical condition first, it is necessary to know what is causing that medical condition. Therefore, in clinical practice, it is often the case that the diagnosis of a medical condition is important by itself and an illustration of this importance can be obtained from the literary fiction. In the Leo Tolstoy’s *The Death of Ivan Ilyich* [10], the drama of a patient who does not know the cause and the prognosis of

his medical condition is brilliantly described and can be synthesized in the following paragraphs of the book:

He said nothing of this, but rose, placed the doctor’s fee on the table, and remarked with a sigh: “We sick people probably often put inappropriate questions. But tell me, in general, is this complaint dangerous, or not?...”

“I have already told you what I consider necessary and proper. The analysis may show something more.” And the doctor bowed.

Ivan Ilyich went out slowly, seated himself disconsolately in his sledge, and drove home. All the way home he was going over what the doctor had said, trying to translate those complicated, obscure, scientific phrases into plain language and find in them an answer to the question: “Is my condition bad? Is it very bad? Or is there as yet nothing much wrong?” And it seemed to him that the meaning of what the doctor had said was that it was very bad. Everything in the streets seemed depressing. The cabmen, the houses, the passers-by, and the shops, were dismal. His ache, this dull gnawing ache that never ceased for a moment, seemed to have acquired a new and more serious significance from the doctor’s dubious remarks. Ivan Ilyich now watched it with a new and oppressive feeling.

In conclusion, although the Fryback and Thornbury 6-step model remains an important and very useful guide in the field of diagnostic test evaluation, we cannot restrict the value of a diagnostic test to situations where it changes the therapeutic process and the outcome of the patients. It is also important that the impact of the diagnosis per se on the patients’ well-being would be considered and the analytical models should evolve to contemplate this essential aspect of the diagnostic process.

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### Methodological studies evaluating evidence are not systematic reviews



When journal editorial staff evaluates submissions of methodological studies in which unit of analysis is a randomized controlled trial (RCT) or a systematic review (SR), they may ask an author to correct study category into an SR and to provide a PRISMA checklist for reporting of SRs. It appears that it is necessary to highlight that methodological studies on RCTs or SRs are not systematic reviews.

After conducting multiple systematic reviews, and not being able to find explicit responses to certain methodological questions, or clear instructions for some methodological steps, I decided to do a number of methodological studies. In those studies, unit of analysis was an RCT or SR. The first such problem I tried to solve is how to extract data from figures. I was conducting a Cochrane review about celecoxib for osteoarthritis [1] and many of my eligible trials reported data only in graphs. I could not find instructions about extracting data from figures in Cochrane Handbook. I tried to find instructions in published Cochrane reviews and, by random search of the Cochrane Library, found only one review where authors described that they printed a figure, magnified it, and then used a pen and ruler to determine value of data points they needed. Then I decided to do a formal study in which I analyzed a number of Cochrane protocols with the aim to see whether authors mention extraction of data figures and which methods they proposed for such extraction [2]. Our team found that only a few of 589 analyzed Cochrane protocols mentioned methods for extracting data from figures. After this initial methodological study, I conducted many more studies in which our team searched

for certain RCTs or SRs, and then we analyzed certain methodological aspects of those studies. In those studies, I usually used certain elements of systematic search or systematic study screening, to find eligible RCTs or SRs.

However, when such studies are submitted to a journal, the editorial staff and reviewers often consider them to be systematic reviews. Editors and peer-reviewers may ask for recategorization of an article from “research” or “original research” into “systematic review.” They ask for revision of the manuscript’s reporting because “a systematic review should be reported according to the PRISMA checklist.” They ask for submission of a PRISMA checklist to make sure that our study is reported in line with requirements for reporting SRs.

Therefore, it should be emphasized that a methodological study that analyzes certain characteristics of evidence is not the same type of study as a systematic review. A systematic review is a study that aims to collect all empirical evidence that fits prespecified inclusion criteria with the aim to answer a specific research question [3]. On the contrary, a methodological study evaluating evidence is aiming to evaluate certain methodological questions and not to collate all empirical evidence on a certain topic. For example, we recently analyzed multiple Cochrane reviews to explore how Cochrane authors analyze risk of bias for included RCTs [4]. Our aim, therefore, was not to collate findings of 721 Cochrane reviews and 10,280 of included RCTs but to analyze certain methodological aspects of that evidence.

In conclusion, editors and reviewers should be cognizant about the existence of methodological studies in the field of evidence synthesis that are not synonymous with systematic reviews. As such, those studies should not be categorized as systematic reviews or requested to be reported as systematic reviews. Such methodological analyses are retrospective cohort studies of certain types of evidence, and not systematic reviews. Systematic searching and screening may be used to identify evidence that will be included, but this does not make a study systematic review.

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