How Case Reports were Defended against Evidence-Based Medicine, The New Empire?

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Editorial

For a few years and since the release of the expression on Medline in 1992 by Gordon Guyatt et al., interest in the Evidence Based Medicine (EBM) had skyrocketed and had induced a kind of globalization of its teaching. Undoubtedly, we live in the era of EBM in which scientific knowledge and research works are classified in a pyramid hierarchy according to their levels of evidence. At the top of this pyramid, level 1, we find randomized clinical trials, meta-analyses, systematic reviews, and secondarily added teleanalysis [1,2]. These studies are expected to have the highest level of evidence. At the bottom of the pyramid, level 4, we find the case reports and case series with a level of evidence obviously lower.

The result of this scientific “segregation” was that case reports began to have low citation rates [3,4], and had become neglected by many medical journals. Case reports became so victims of a real censorship with the famous impact factor as the main agent. This new editorial policy particularly of many prestigious journals had led to a serious doubt on the interest of case reports and authors and readers had begun to wonder if they should always take the time to write or read new cases. Taking advantage of the situation, some “statisticophiles” began to make statements of “case-reportophobia” like “case reports do more harm than good by emphasizing the strange and the bizarre” [5] ... And sometimes we could hear arrogant and contemptuous statements like “Case reports are the back water for old fogies and experts of the exotic” [6]. Why this ungrateful attitude toward a secular cornerstone of the medical literature, the case report? Why many authors and readers had quickly succumbed to unfair new requirements of the modern medicine, the EBM?

Being aware of the danger threatening the case report, many old hands of writing had revolted and came to its defense. Why? Simply, because the case reports will always have something to offer to medical literature. Richard Smith, former editor-in-chief of BMJ (British Medical Journal), a journal with a high impact factor, expressed his desire in 2008 to be editor-in-chief of a case report journal [7]. In his book “Clinical Case Reporting in Evidence Based Medicine”, Milos Jenicek [8] defended case reports and stated “The case reports and cases series may have the lowest evidence level, but they are often the first line of evidence ... This is where it all begins”. Yes, the beginning of medicine is always an event and its history is strongly linked to case reports. In 2014, Andrew Lane [9] titled an editorial about case reports: “The case report: Level 5 evidence, Level 1 medicine”; this very expressive title sums up everything.

The case report had then to adapt to novel scientific requirements and its purpose had evolved from simple reporting rare or unusual conditions to pragmatic help of clinicians in diagnosis and treatment of less common diseases [10]. Furthermore, some authors [8,11] demonstrated how to apply concepts, principles and statistical methods of clinical trials and systematic reviews to the management of individual clinical situations and a new concept of “evidence based case report” was then developed.

New peer-reviewed journals publishing case reports quickly emerged over the last several years. In 2015, there were 160 journals from 78 publishers with nearly a half indexed in PubMed [12]. Moreover, to increase the completeness and transparency of published case reports, guidelines have been developed (CARE) as for randomized controlled trials (CONSORT), and systematic reviews and meta-analyses (PRISMA) [13].

With a vision that case reports would have much more value if they could be assembled together in large numbers and made easy to find and compare, BioMed Central (the online publisher of free
peers-reviewed scientific articles) had launched on December 2012, Cases Database, a continuously updated and free-accessible resource [14].

It is not the end of this provoked defense movement. In fact, international conferences dedicated for case reports, are currently frequently being organized (http://clinicalcasereports. conferenceseries.com). In addition, several web communities have developed for the discussion of case reports like DG Cases (http://dgcases.docguide.com) and Orthogate Cases (https://www.orthogate.org/cases).

No one can deny the contribution of EBM in doctors’ decisions and no one can ignore the scientific merit of the case report. There is no doubt that the case reports cannot replace the EBM. But there are gray areas in medicine where certain diseases remain incognito and where EBM with its powerful and complex mechanisms, cannot enter. The statistical and clinical epidemiology is not adapted to meet the specific issues at individual level.

Let us now remember some interesting stories of case reports merits.

In 1999, the use of a series cases in the recognition of a new disease was illustrated by the epidemic of West Nile encephalitis in New York. Indeed, the West Nile virus has never been reported before in the Americas. But the astute observation of a small number of cases in humans and crows living freely in the zoo had allowed to detect virus and identified a possible transmission route between human and birds [15]. The outbreak was then followed by a spectacular spray operation of insecticide (Malathion), on certain parts of the city by helicopters, an equivalent of the Clean Sweep Operation that Colonel Sam (Dustin Hoffman) had aborted in "OUTBREAK", the film.

Case reports are also important for the detection of side effects of new drugs, whether negative or positive. In fact, they were the direct cause of a serial withdrawals of drugs; poor Rofecoxib (VIOXX)!!! Conversely, Sildenafil (VIAGRA) savior face of some of us had been developed from fortuitous observation of a side effect of antihypertensive treatment [8].

The case reports or the anecdotes in medicine as like to call them certain authors had also inspired screen writers and film and television producers. In 2004, David Shore and Paul Attanasio, pitched the television series titled Dr House to Fox television. It was inspired from The New York Times Magazine column, "Diagnosis", written by physician Lisa Sanders, which discusses rare cases that baffled medical teams. House was the most-watched television program in the world in 2008 [14].

Case reports have and will continue to have their place in the literature despite their poor ranking in the hierarchy of evidence [16]. With the promotion of the role of case report through ANNCASEREP,

we hope to participate in building a large database of case reports. This database, a true Case Bank, will be a source allowing researchers to get answers to specific questions about a rare event for example. Marta Herreros-Villanueva [17] reviewed the most important cases of spontaneous regression of pancreatic cancer that have been reported in the literature and discussed whether it is a real phenomenon or a misdiagnosis.

References