

# In the land of double-blind studies the case report is king

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Scientific journals publish a myriad of various articles, of which reports of original scientific findings and review articles often represent the backbone of the journal. In addition, opinion-based articles, hypothesis papers, editorials, letters and rebuttals, news and views, and many other article types may be published. A particular type of paper that is typically seen in medical journals only is the case report. The case report is a popular item for many medical authors and represents about 10-15% of indexed papers in PubMed. In a case report doctors report about their observations in a particular patient or a small series of patients. Reasons for publishing a case report may vary but include the situation in which doctors are impressed by a certain patient presentation and/or clinical course, the authors' conviction that colleagues need to be informed about a specific clinical situation or development, and initial reports on new insights into pathogenesis, a novel diagnostic technique or an innovative therapeutic option. Also the case reports published in the Netherlands Journal of Medicine fall into these categories. For example, in recent years we have published interesting new aetiological or pathophysiological findings,<sup>1,2</sup> new diagnostic or laboratory techniques,<sup>3</sup> original clinical manifestations of diseases,<sup>4,5</sup> or new adverse events of treatment.<sup>6-8</sup>

There are also a number of downsides to case reports that are frequently mentioned. In the first place, case reports are anecdotes and illustrations, and their narratives may represent coincidences rather than a real significant trend that helps in understanding disease or improving diagnostic or therapeutic management. In particular case reports describing the coincidental occurrence of two different diseases are not very helpful. If an individual patient with chronic ulcerative colitis develops glioblastoma multiforme, it is quite likely that this is a coincidence rather than pointing to a genuine connection between the two diseases. Nevertheless, case reports like this are very often submitted to journals and sometimes even published. Secondly, there is a marked publication bias associated

with case reports: only impressive or interesting situations are likely to be reported. Lastly, the lack of a control group or situation makes it very hard to adequately assess the true importance and relevance of any individual observation. As an example, the publication history of a recombinant activated coagulation factor to combat massive blood loss can be taken. This intervention showed highly impressive and almost immediate effects in arresting blood loss in some patients with uncontrollable haemorrhage in whom all other options had failed. The medical literature was barraged with an extreme number of case reports and case series reporting successful application of this treatment. Patients in whom this therapy failed were almost never reported, underscoring the publication bias this type of articles may have. For many years the number of patients reported in case reports exceeded the number of patients included in clinical trials and for a long time this has hampered a proper assessment of the true efficacy and safety of this intervention.<sup>9</sup>

Despite these disadvantages some authors argue that case reports can also have merit in some areas. Vandembroucke argues that case reports, when clearly focused, are often crucial in detecting novelty and may therefore be instrumental in stimulating medical progress.<sup>10</sup> This notion was confirmed by another report demonstrating that novel observations in case reports were often followed up by subsequent clinical trials and could therefore be considered to be important hypothesis-generating reports.<sup>11</sup> The undiminished popularity of case reports is underlined by the number of submissions to the Netherlands Journal of Medicine (*table 1*) and this ever increasing number of submissions does lead to a decreasing chance of acceptance.<sup>12</sup> In the Journal the editorial policy is to be restrictive about case reports (and basically limit acceptance to those cases that really report novel ideas or findings) and to publish interesting and illustrative examples of disease in the photo quiz section. In fact, the photo quizzes are highly popular items in the Journal and

**Table 1.** Number of submissions and acceptance rate of case reports and photo quizzes in the Netherlands Journal of Medicine

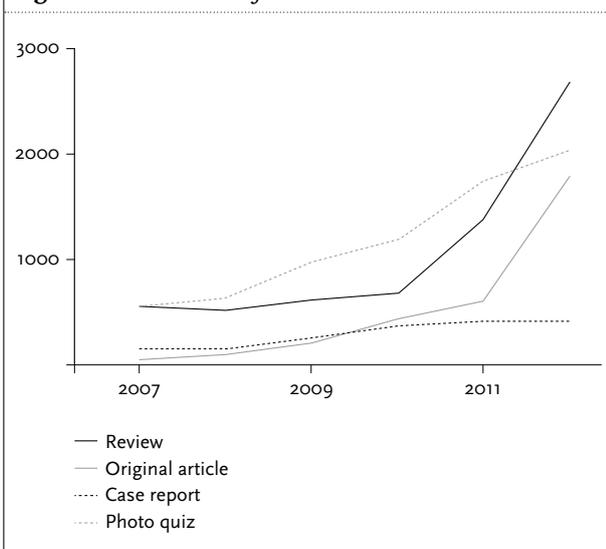
Year	Submitted case reports	Accepted case reports	Submitted photo quizzes	Accepted photo quizzes
2009	136	14%	50	68%
2010	272	11%	71	56%
2011	284	9%	63	63%
2012	372	7%	141	35%

**Table 2.** Most frequently downloaded photo quizzes in the Netherlands Journal of Medicine

Photo quiz	Number of downloads*
Smit TT, et al. An odd looking man <sup>13</sup>	2714
Martens H, et al. A 'chigsaw' puzzle after a vacation in Brazil <sup>14</sup>	2408
Tummers-de Lind, et al. Nodules on the tongue and thick lips <sup>15</sup>	2298
Lu H, et al. Pythons and a palmar rash <sup>16</sup>	2282
van Durme CM, et al. Dripping candle wax <sup>17</sup>	2168

\*Downloads within one year of publication.

**Figure 1.** Downloads of articles



Annual mean number of downloads of several article types in the Netherlands Journal of Medicine from the Journal's website (open access).

belong to the most frequently downloaded articles on our website (figure 1). The five most frequently downloaded photo quizzes are shown in table 2.

The case report is often regarded as a less valuable contribution to the medical literature; however, it is still widely popular among authors and probably also among readers. Hence, this publication form is likely to stay in our journals for many years to come.

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