

# Chapter 2

## The Historical Tradition of Case Reporting

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### The Case Report in Ancient and Medieval Times

The medical case report as we know it today is not a static form of medical communication. It has evolved over almost 4000 years, and the format, content, and uses of case reports have undergone remarkable changes according to the shifting historical, technological, and cultural contexts. While the tradition of case reporting is ancient, our current way of writing a case report is a relatively recent development.

The first known medical case reports, circa 1600 BC, were written and preserved on an Egyptian papyrus. They comprise a series of 48 cases which discuss injuries and disorders of the head and upper torso, and include an accurate description of a maneuver to reduce a jaw dislocation [1]. Another ancient Egyptian medical treatise, the Ebers papyrus (1552 BC), is a 110-page scroll which contains folk remedies, magical potions, and descriptions of a wide variety of diseases, including a disorder of frequent urination that is probably the first report of diabetes mellitus (although an ancient Hindu text from the same period noted that ants were attracted to the urine of people with a mysterious wasting disease) [2]. Egyptian medicine was practiced by physician-priests, and treatment, though often practical and occasionally useful, was inextricably

entwined with magic [3]. In contrast, the Hippocratic case histories from the Greek classical era (ca. 400 BC) show no belief in supernatural origins of disease, and are characterized by objective and detailed descriptions of the findings and courses of various illnesses [1]. The physician-narrator was generally an observer, and did not intervene or otherwise participate in the case [4]. This case history from the *Epidemics*, of a patient with what sounds like a foot infection complicated by cellulitis and overwhelming sepsis, is typical:

Criton, in Thasus, while still on foot, and going about, was seized with a violent pain in the great toe; he took to bed the same day, had rigors and nausea, recovered his heat slightly, at night was delirious. On the second, swelling of the whole foot, and about the ankle erythema, with distention, and small bullae (phlyctenae); acute fever; he became furiously deranged; alvine discharges bilious, unmixed, and rather frequent. He died on the second day from the commencement. [5]

Hippocrates' crowning achievement was the *Aphorisms*, a collection of 412 short, pithy maxims which conveyed the collected "teaching points" of his case histories. Aphorisms lend themselves to oral transmission – they live in speech, not on the page – and the best and most memorable of them are like poetry, the haiku of medicine. "It is the oral-poetic nature of a good aphorism," writes Quentin Shaw, "that flash-welds it into the memory" [6]. The aphorism has persisted as a teaching tool; consider three Hippocratic aphorisms [7], followed by three that I memorized from my own medical training:

Dysentery, if it commence with black bile, is mortal.  
 In dropsical persons, ulcers forming on the body are not easily healed.  
 In acute diseases, coldness of the extremities is bad.  
 The pain of biliary colic radiates to a point just below the right scapula.  
 The sun should never set on an empyema.  
 Chest pain that is substernal, exertional, and relieved by nitroglycerine is angina pectoris.

In the modern case report, it is still customary to conclude the discussion with an aphoristic teaching point. As we sum up the case, we invoke the authority of Hippocrates.

Galen (129–ca.200 AD), the eminent Greek physician and philosopher, took a very different approach to the case history. In contrast to the objective and detached case descriptions of Hippocrates, Galen used the first person to place himself at the center of his cases. His subjects were frequently celebrities – senators, philosophers, even the emperor – and he did not hesitate to describe his brilliance as a diagnostician, the gratitude of his patients, and the shortcomings of other physicians. Regarding Galen's first work, *On the Affected Parts*, Cristina Alvarez Millan comments:

...we have in these accounts mainly strange conditions and spectacular cures, and more importantly, we also find the literary device of increasing suspense throughout the nosological description, together with a remarkable amount of theatrical rhetoric. Thus, a typical clinical account would be in the following sequence: Galen's fear for the patient, Galen questioning the patient, Galen puzzling over the disease, Galen coming to a conclusion (diagnosis or prognosis), Galen warning the patient, Galen preventing his colleagues from administering a certain treatment, Galen amazing everybody on the scene, Galen explaining the real nature of the matter, and so on. [8]

Galen made many brilliant observations, and his medical teachings were revered for more than a thousand years after his death, until Vesalius and others began to disprove his anatomical and physiological theories.

In the middle ages, European medical progress was hampered by rampant scholasticism and religious proscriptions against dissection and experimentation. As the Dark Ages continued in Europe, Islamic medicine took the lead, most impressively in the work and writings of Rhazes (865–925 AD) and Avicenna (980–1037 AD). The case histories of Rhazes stand out in particular for their accurate and insightful descriptions of disease. Rhazes was the first to differentiate smallpox from measles, and his explanation of why survivors of smallpox are not infected a second time stands as the first theory of acquired immunity [9]. In another case, of a man who had fallen from a horse and sustained a neck injury with loss of sensation in his third, fourth, and fifth fingers, Rhazes was able to localize the lesion to “the nerve

located after the seventh vertebra” (i.e., C-8) [1]. Rhazes had many students, and clearly used his case reports as didactic tools; in many instances, he went beyond description to include his own comparisons and generalizations from previous cases [10].

There was an interesting parallel rise of the case report in both Europe and China in the late fifteenth century. The first European collection of cases came from the Portuguese Jewish physician Amatus Lusitanus, who published 700 cases from 1551 to 1556. In China, the *Stone Mountain Medical Case Histories of Wang Ji*, written up and published in 1531 by his disciple Chen Hue, comprised about 100 case histories collected over 15 years. The early Chinese and European case reports had similar formats, and served both to teach a wide audience and to promote the doctors’ practices. An important difference was that the Chinese case reports tended to put respect for authority above experience, while the European case reports emphasized experience and valued debate and argument. Also, the Chinese physicians thought that case reports were for patients as well as physicians, and functioned as part of the treatment; European physicians saw them chiefly as a way to communicate with each other about diseases and treatments [11].

## Origins of the Modern Case Report

The historian Gianna Pomata has traced the roots of the modern case report to the *Observationes* (collections of case histories), which originated in the second half of the sixteenth century, and grew to become a primary form of medical writing by the eighteenth century [12]. The *Observationes* began as a form of self-promotion for town and court physicians, but eventually came to be viewed as a source of medical knowledge. “The original emphasis on therapeutic success,” writes Pomata, “gave way to a new focus on the descriptive knowledge of disease through detailed observation.”

As the Enlightenment eased religious restrictions, autopsy findings began to be included in case reports, which improved diagnostic accuracy and led to major advances in the study of anatomy and physiology [13]. Pomata links the rise of the case report with “the new epistemological value of observation in the age of the Scientific Revolution” [12]. From the eighteenth century to the mid-twentieth century, even as it continued to change and develop, the case report played a critical role in teaching and discovery as well as in shaping the day-to-day practices of physicians. This is not to say that the case reports of 200 years ago were much like the objective and detached reports of today; eighteenth and early nineteenth century authors still favored the conversational tone of Galen, placed more emphasis on the patient’s subjective experiences, and often employed “dramatic devices” to delay the moment of diagnosis or heighten the narrative tension [4]. Consider Dr. John Warren’s description of a case in his *Remarks on Angina Pectoris* (1812):

I had too soon an opportunity of confirming my suspicions; for on the following Sunday, whilst attending public worship in Brattle Street, Mr. Neal was seized with a most violent paroxysm, under circumstances peculiarly affecting. In the midst of a discourse highly interesting in its nature, and delivered with a great degree of fervor, whilst the eyes of all were fixed upon the preacher, he was observed to raise his hand, and forcibly rub his breast; his voice faltered, and his countenance changed; and, after one or two efforts to proceed, he sallied back on his seat, and became insensible. [14]

In his *Essay on the Shaking Palsy* (1817), James Parkinson brilliantly linked the disorders of trembling, posture, and gait as a single disease entity. Here he describes the terminal stages of the disease:

As the debility increases and the influence of the will over the muscles fades away, the tremulous agitation becomes more vehement. It now seldom leaves him for a moment; but even when exhausted nature seizes a small portion of sleep, the motion becomes so violent as not only to shake the bed-hangings, but

even the floor and sashes of the room. The chin is now almost immoveably bent down upon the sternum. The slops with which he is attempted to be fed, with the saliva, are continually trickling from the mouth. The power of articulation is lost. The urine and fæces are passed involuntarily; and at the last, constant sleepiness, with slight delirium, and other marks of extreme exhaustion, announce the wished-for release. [15]

Note the personification (“exhausted nature”) and the sentimental “wished-for release” which further dramatize Parkinson’s compelling clinical observations. In a similar vein, several eminent Victorian cardiologists used sensational and sentimental language in their case reports as they described the distressing effects of heart disease on their patients – and themselves [16].

But change was coming: the twentieth century saw the depersonalization of case reports and the standardization of their structure, with the rise of the now-familiar “introduction/case report/discussion” format and the gradual disappearance of the author from the narrative [1]. Osler’s 1902 report of two cases of intermittent claudication [17] is characteristic of this modern transition point in the case report. He begins with a recollection of a horse autopsy he had viewed with some members of the Montreal Veterinary College more than 20 years before. The horse had been afflicted with a “peculiar form of intermittent lameness,” and the autopsy showed “verminous aneurysms...of the iliac arteries.” He cites the case of another horse that had to stop and rest “after being driven for fifteen or twenty minutes;” autopsy showed clots obstructing the arteries in both hind legs. He then gives a thorough review of the literature, including a case reported by Charcot in 1856 of a soldier with classic intermittent claudication, who was found at autopsy to have a bullet encysted near the iliac artery, which had caused an aneurysm with obliteration of the lower part of the artery. Collateral blood flow had allowed a modest level of activity, but more vigorous activity caused ischemic pain that was relieved only with rest. Osler then describes his own case of a young man with a

syphilitic abdominal aortic aneurysm who developed leg claudication symptoms after the aneurysm was successfully treated with “wiring and electrolysis”:

...After walking for a certain distance his legs would, as he expressed it, give out completely; so that he could not move another step, and had to sit down. After resting a few minutes he could then go on again. This was more particularly noticeable when he walked on the street. He had to go very slowly and could not go for any distance. There was no paralysis accompanying the loss of ability to walk. He could move his legs, but there was an uncontrollable feeling that he could not take another step. Accompanying this there was a sensation of dead, heavy weight in the legs, but no cramps. Walking about in the house (and in the yard) did not bring on the condition, but he had had it very frequently in the past few months and he had learned to ward it off by walking very cautiously and slowly and resting at intervals. The femoral artery and the dorsal arteries of the feet were distinctly sclerotic.

Osler concludes with an important teaching point:

As shown in the horse and in the first case which I here report, the affection is not always due to simple arterio-sclerosis, but may be due to aneurism, as in Charcot's case and as is the rule in the horse. [17]

In this terrifically interesting and entertaining interspecies case report, Osler writes in the first person, and brings in his odd (though very apt) experience in veterinary medicine. Yet, this case report is distinctly modern in that it contains an introduction, review of the literature, concise description of two cases, and a strong teaching point. At the end, Osler cannot resist adding “a word as to the name,” and indicates his preference for the term “*intermittent claudication*” as opposed to *angiosclerotic intermittent dysbasia*, *intermittent muscle paresis*, or *angiosclerotic paroxysmal myasthenia*, as proposed by Charcot, Erb, and Higier. This is the literary Osler expressing a preference, which any reader of *Aequanimitas* will surely respect. (Osler's paper brings to mind another interspecies case report, recently published in the *New England Journal of*

*Medicine*, of an HIV-infected man who developed a nonhuman malignancy that arose from proliferating, genetically altered tapeworm cells. This was the first case of human disease caused by transmissible clones of parasite-derived cancer cells [18]).

## The Rise, Decline, and (Electronic) Rebirth of the Case Report

Over the past 100 years, the popularity of the case report has risen, fallen, and risen again. The twentieth century saw a tremendous surge in the publication of case reports focusing on new diseases, drug side effects, etiology and mechanisms of disease, therapy, prognosis, and education [19]. New diseases first described in case reports include shell shock (1915), Cushing's syndrome (1932), erythroblastosis fetalis (1932), Ebola virus infection (1977), toxic shock syndrome (1978), AIDS (1981), and thrombophilia due to Factor V Leiden (1993). Significant drug side effects first described in case reports include thalidomide-related birth defects (1961), venous thrombosis due to oral contraceptives (1961), chlorpropamide-induced SIADH (1970), valvulopathy associated with weight-loss drugs (1996), and troglitazone-induced liver failure (1998). Other landmark twentieth century case reports include the first surgical ligation of a patent ductus arteriosus (1939), the first use of lithium to treat mania (1949), and the first heart transplant (1967). Physicians looked to case reports for practical guidance, education, and inspiration. Furthermore, they were frequently able to publish their own interesting cases; over the 30-year period, from 1946 to 1976, case reports comprised up to 38 % of all articles published in general medicine journals [20].

The 1980s, however, marked the beginning of a steep decline in the publication of case reports in many leading journals. This decline correlated with a rise in the publication of "research articles" (articles which included original,

firsthand data with a clearly delineated research methodology), which increased from 50 % of all psychiatry journal articles in 1969–1970 to 82.4 % by 1989–1990 [21]. Another factor was the popularization and broad acceptance of “evidence-based medicine,” which raised randomized controlled trials to the pinnacle of the evidence pyramid, and vilified case reports and case series as “the lowest forms of intellectual life, even lower than the case-control study” [19]. In their excellent article on the recent history of the clinical case report, Nissen and Wynn [22] describe how the case report was marginalized by editors, whether relegated to the “Letters to the Editor” section, limited by stringent selection criteria, or, in many cases, barred entirely from publication. Case reports are generally cited less often than clinical research studies, and the increasing importance of the impact factor gave editors another reason to avoid publishing them. In addition, the dramatic rise in government and pharmaceutical company funding of randomized controlled trials made the largely unfunded case report a less desirable venue for both authors and journals [22]. All of these factors contributed to the flat overall growth in case report publication from the 1980s through the mid-1990s.

And then, in the late 1990s, the case report began to rise again. A number of prestigious journals – *The Lancet*, the *American Journal of Psychiatry*, *BMJ*, the *Journal of Clinical Oncology*, and others – began to publish case reports again in a variety of formats. Around the same time, some skeptics began to criticize evidence-based medicine as “a hierarchy of clinical epidemiology” that favors large-*n* quantitative studies to evaluate medical interventions with wide applicability, but fails at the level of the individual patient [22]. In the backlash against evidence-based medicine, interest began to grow in the narrative of the individual patient, which could be studied with qualitative research methods and applied to other patients with similar patterns of illness. Between 2000 and 2013, the rate of PubMed-indexed case report publication increased by 36 %, from 42,000 to 58,000 per year.

Perhaps the most important factor in the case report's revival has been the rise of the electronic case report journal. This trend began in 2007, with the *Journal of Medical Case Reports*, *Cases Journal*, and *BMJ Case Reports*; since then, there has been exponential growth with more than 30 online case report journals as of November, 2015. Increasingly, both the advantages and problems of these online journals are becoming more clear [23]. On the one hand, the case report journal can be a gateway to publication and career advancement for both busy clinicians and novices who lack experience in medical writing. In areas of the world with limited resources, where research funding is scarce, open-access case report journals can be a way to post important clinical findings and circulate medical information in the community. Another potential benefit is the creation of a large database of cases, which in theory could be useful in the diagnosis and treatment of individuals with complex and unusual problems not addressed by clinical trials. On the other hand, the quality of both peer review and writing has been inconsistent in some of these journals, with critical information missing, inadequate explanation of events, and unsupported conclusions. Authorship fees can be a hindrance to publication, especially for authors lacking institutional support, and the combination of high fees and substandard peer review raises the specter of "vanity publication" for those willing to pay. Finally, the large database of marginally significant cases in online journals "might drown key sentinel events in a sea of careless publishing" [23].

Several authors have proposed case report guidelines as a solution to these problems [23–25]. They suggest that guidelines could improve both the evidence value of individual case reports and the quality of the database as a whole. The authors of the CARE guidelines, a consensus-based case report guideline and 13-point checklist first published in 2013, assert that "the systematic aggregation of information from case reports will inform clinical study design, provide early signals of effectiveness and harms, and improve health-care delivery" [22]. This is an exciting development, but one

could argue that, with 1.77 million case reports already indexed in PubMed, the guideline “ship” has already sailed. Another approach might be to disseminate the CARE guidelines to all physicians in training and teach them to assess the evidence value of each case report at the time it is accessed for patient care.

But before we embrace strict guidelines, we should consider what we have learned from the long history of the medical case report. Case reports reflect the cultures, values, and technologies of their times. Therefore, the form and function of the case report must continue to change over time. Who can imagine what medical case reports will look like in 500 years? And why would these future case reports adhere to twenty first century guidelines, any more than we look to the sixteenth century for instruction? The other distinctive thing about case reports is that they are at their best highly creative endeavors, bursting with enthusiasms, intuitions, and hypotheses that are barely restrained by the traditional form. Is there a place within the guidelines for Osler’s stiff-legged horses, or for Rhazes’ intuitive hypothesis on immunity, which was correct but remained unverifiable for centuries? Whatever guidelines we choose, we must not stifle speculation, which is essential not only in case reporting but in all medical writing.

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