



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Editorial

Time for a new approach to case reports

Case reports are popular and represent one of the oldest levels of evidence. Despite this they have attracted much criticism. They can be trivial, their conclusions do not usually undergo further investigation, and they are often questioned as a poor excuse for an unstructured review of the literature. Lessons from the past have given strong warnings against using new drugs based solely on case reports, such as the long-term adverse consequences of fetal exposure to diethylstilboestrol (DES) in pregnancy.¹ Case reports are often not cited and hence many journals routinely reject them to protect their impact factors. Indeed, there is evidence that the prevalence of these articles in high impact journals is in decline.²

Many journals have reconsidered their policy towards case reports over the years^{3,4} and there has been a recent vogue in new journals publishing exclusively case reports,^{5–7} one of which is no longer active.⁸ The *International Journal of Surgery* (IJS) has had dissonance towards case reports since its launch in 2003. Numbers steadily rose until they formed over a quarter of submissions in 2007. Under the weight of these submissions we chose to institute a policy of non-acceptance in late 2007, our instructions to authors were updated and many enthusiastic authors were rejected. Despite this, case reports have continued to be submitted in large numbers. We tentatively started to accept case reports again in mid-2009 with the bar set very high on quality and novelty. We feel the time has now come for the first dedicated home for such articles – *International Journal of Surgery: Case Reports (IJSCR)*. The journal is due for launch on 20 May 2010 and the IJS itself will no longer accept case reports from this point forward. This editorial provides a synoptic exposition for this major change in direction and its underlying rationale.

Clinicians – and surgeons in particular – learn from stories, anecdotes and patient-based learning. Such learning has been essential to the advance of healthcare.⁹ Clinical knowledge is remembered far more efficiently when tagged to real-life events; this is perhaps to be expected amongst human beings working in such a highly organised social environment. Case reports can be very sensitive at detecting novelty. By definition, the discovery of every new condition, be it AIDS, SARS, Swine Flu or the next emergent disease begins with a single case. They complement evidence-based medicine, the learning of which should be drawn from a broad range of sources.^{10,11} A recent study of drugs withdrawn from the market found the scientific evidence for 19 out of 21 drugs came from spontaneous case reports. Case reports were the sole evidence in 12 and a randomised controlled trial (RCT) was only responsible for one.^{12–15} Aronson¹⁶ and Glasziou et al.¹⁷ have described circumstances where case reports have provided definitive not just indicative evidence. Individual cases can also have a major impact

on health policy. In the UK, on 4 February 2010, Coroner William Morris ruled the death of a 70-year-old patient who was administered a tenfold overdose of diamorphine by a German GP was an unlawful killing.¹⁸ This resulted in prompt action by the UK Government to prevent similar cases.¹⁹

The relatively 'open' format of case reports for discussing atypical presentations, diagnostic challenges/pitfalls and rare conditions is especially useful. Their less rigid structure allows the surgeon(s) to discuss their diagnostic approach, the context, background, decision-making, reasoning and outcomes. Their human perspective would be of unique and special value and provide insights into the doctor–patient relationship. In addition, by writing about their experiences, surgeons gain the opportunity to reflect on their practice, an important aspect of personal development often not afforded the time amongst busy clinics and theatre lists. Yet we know their intrinsic value from patient safety incidents, root cause analysis, morbidity and mortality meetings.

Case reports may remain the 'lowest' or 'weakest' level of evidence with respect to causality but they remain the first line of evidence of what actually happened. They allow for the demonstration of novel surgical techniques (as laparoscopic cholecystectomy was once upon a time) prior to more substantial comparative analysis with established techniques such as RCTs. The need for more case reports may have been paradoxically increased by the drive for big RCTs. One of us (RA) has already demonstrated some of the problems with surgical RCTs.²⁰ People with three or more chronic conditions account for around 80% of healthcare activity; however, the evidence base that relates to such people is poor. RCTs tend to exclude patients who have more one than chronic condition in order to limit the number of variables, potential confounding factors and to aid interpretation. Yet how many of us deal with patients who may have two or more of the following: diabetes, arthritis, heart failure, depression, chronic obstructive airways disease, asthma and so on. This is not uncommon, 40% of patients with long-term conditions have more than one condition, and many have three or more.²¹ UK National Institute for Health and Clinical Excellence (NICE) guidelines are based on trials that deliberately excluded patients with comorbidities. You just cannot do RCTs on every kind of patient but these are the types of patients we see day in day out. The importance of not forgetting outliers has been discussed elsewhere.²² Outliers in larger studies are statistically sidelined and further analysis of unexplained and unexpected observations is not conducted. It is therefore hard for such patients and their surgeon to know what's best.

The other issue is generalisability. RCTs are often conducted in centres of excellence with the necessary funding, support and

structure to make them happen. Their results may not be easily extrapolated to low- and middle-income countries with limited resources. RCT evidence on children, tropical diseases and surgical conditions not found in the developed world is particularly poor.²³ Searching our database for patients in similar circumstances treated to the best possible standards locally in the health service structure and cost-base of that country would provide value and aid the decision-making process for both surgeon and patient. Such reports stimulate learning and research and are capable of developing new subject areas as well as providing excellent educational material to students, trainees and lecturers. The addition of multimedia facilities such as online video is of particular relevance to surgery – a craft specialty.

In recent times, the first face transplant²⁴ and the first uterine transplant²⁵ were presented as single cases. Despite the technical successes and failures of such attempts at novel transplantations, the reports themselves allowed the international scientific and surgical community to gain useful insights at the 'cutting edge'. We learnt about the technical difficulties, immunological issues, psychosocial and ethical dilemmas from the 'front line' so to speak. It is with the richness of such cases and demand from authors that we feel the time has come to launch IJSCR.

IJSCR will be online-only, peer reviewed and will only publish case reports which are clinically interesting, original and educational. All published content will be universally available to everyone on our dedicated website www.casereports.com and the Science Direct platform (www.sciencedirect.com), which is one of the world's most advanced web delivery systems for scientific, medical and technical content hosting more than 10 million articles with more than 11 million users. All published content will be publicly posted on PubMedCentral (PMC) and our aim is to ultimately achieve indexing in Medline as well. The journal will also be indexed in SCOPUS, EMBASE and Google Scholar soon after launch on 20 May. This business model of freely accessible content will necessitate all authors paying an article processing fee of £250 (plus VAT/Sales tax) should their article be accepted for publication. This is in keeping with other journals that operate such a business model²⁶ and compares favourably.

Types of cases we will be interested in include:

1. Reminder of an important clinical lesson.
2. 'How I do it'.
3. Findings that shed new light on the possible pathogenesis of a disease or an adverse effect.
4. Learning from errors.
5. Unusual presentation of more common disease/injury.
6. Myth exploded.
7. Rare disease.
8. New disease.
9. Novel diagnostic procedure.
10. Novel treatment (new drug/intervention, established drug/procedure in new situation, use of new technology).
11. Unusual association of diseases/symptoms.
12. Unexpected outcome (positive or negative) including adverse drug reactions.

Cases will be judged on clinical interest and educational value not novelty or rarity. All authors should refer to our Guide to Authors available online in the first instance (<http://ees.elsevier.com/ijscasereports>). We expect case reports to be short and simple to prepare, thus providing an ideal way trainees to gain writing and publishing experience.

This new journal in no way detracts from more rigorous studies which the IJS will continue to publish and we wish to echo Sir Iain Chalmers' comments²⁷ in response to the launch of another case

reports journal:

"In 1952, Austin Bradford Hill – the medical statistician who play such an important part in introducing the randomized controlled trial to medical research – 'In my indictment of the statistician, I would argue that he may tend to be a trifle too scornful of the clinical judgments, the clinical impression. Such judgments are, I believe, in essence, statistical;. The clinician is attempting to make a comparison between the situation that faces him at the moment and a mentally recorded but otherwise untabulated past experience'. Twenty years later, Sam Shuster – a clinician – warned that these impressions can be seriously misleading: 'There are lies, damned lies, and clinical impressions'²⁸. Both Bradford Hill and Shuster are right, of course: informal evaluation of care based on impressions, and formal evaluation based on well-controlled comparisons of alternative forms of care, both play essential roles in the promotion of more effective care. . ."

We aim to develop the science of case reports and raise their academic value. Ultimately, these case reports will form a rich database, a corpus of clinical information which could be mined for interesting trends/patterns, disease modelling and for the purposes of surveillance (especially of rare diseases). Examples of such collective value internationally include:

- The Global Trigger Tool from the Institute for Healthcare Improvement (IHI) in the USA which has been shown to increase the rate of adverse drug event detection approximately 50-fold over traditional reporting methodologies.²⁹
- The Medicines and Healthcare products Regulatory Agency's (MHRA) YellowCard³⁰ reporting system for adverse drug reactions (ADRs).
- The National Patient Safety Agency (NPSA) in the UK, whose National Learning and Reporting System³¹ (NLRS) relies on single cases and where extraction of systemic lessons from such data is now beginning to bear fruit.³²
- The BEACH (Bettering the Evaluation And Care of Health) programme in Australia has used 100 consecutive and randomly sampled case reports each year from general practitioners to describe 'the characteristics of GPs and the patients who consult them, patient reasons for encounter, the problems managed and management techniques used'. The programme now has over 10 years of data and has been very important in developing general practice in Australia.³³

We hope that our database will be greater than the sum of its parts too and the 'wisdom of crowds' will take it to new heights.³⁴ This editorial represents an open call to the Sherlock Holmes within us all and to the wider surgical community. Join us in this exciting new venture and demonstrate the rich diversity of clinical surgery by submitting your best cases to IJSCR at: <http://ees.elsevier.com/ijscasereports>.

References

1. Chalmers I. Evaluating the effects of care during pregnancy and childbirth. In: Chalmers I, Enkin M, Keirse MJNC, editors. *Effective care in pregnancy and childbirth*. Oxford: Oxford University Press; 1989. p. 3–38.
2. Mason RA. The case report – an endangered species? *Anaesthesia* 2001;**56**(2):99–102.
3. Bignall J, Horton R. Learning from stories – the Lancet's case reports. *The Lancet* 1995;**346**(11):1246.
4. Benninger MS. The value of case reports – our journal's approach. *Otolaryngology – Head and Neck Surgery* 2005;**133**:1–2.
5. Jenkins D. What shall we do with case reports? *British Medical Journal Group Blogs* 14 November 2008.
6. Kidd M, Hubbard C. Introducing journal of medical case reports. *Journal of Medical Case Reports* 2007;**1**:1–2.
7. Smith R. Why do we need cases journal? *Cases Journal* 2008;**1**:1–2.

8. Smith R. Cases journal: time for a new path. *Cases Journal* 2009;**2**:9122.
9. Jenicek M. *Clinical case reporting in evidence-based medicine*. 2nd ed. London: Arnold; 2001.
10. Agha R, Singh G. Studying for an Intercalated BSc? *Medical Education* 2003;**37**(9):839.
11. Greenhalgh T. Doing an intercalated BSc can make you a better doctor. *Medical Education* 2003;**37**:760–1.
12. Vandembroucke JP. In defense of case reports and case series. *Annals of Internal Medicine* 2001;**134**:330–4.
13. Vandembroucke JP. Case reports of suspected adverse drug reactions: case reports were dismissed too quickly. *British Medical Journal* 2006;**332**:488.
14. Aronson JK. Anecdotes as evidence. *British Medical Journal* 2003;**326**:1346.
15. Loke YK, Price D, Derry S, Aronson JK. Case reports of suspected adverse drug reactions – systematic literature survey of follow-up. *British Medical Journal* 2006;**332**:335–9.
16. Aronson JK, Hauben M. Anecdotes that provide definitive evidence. *British Medical Journal* 2006;**333**:1267–9.
17. Glasziou P, Chalmers I, Rawlins M, McCulloch P. When are randomised trials unnecessary? Picking signal from noise. *British Medical Journal* 2007;**334**:349–51.
18. Meikle J, Campbell D. Doctor Daniel Ubani unlawfully killed overdose patient. *Guardian.co.uk*. Available at: <http://www.guardian.co.uk/society/2010/feb/04/doctor-daniel-ubani-unlawfully-killed-patient> [accessed April 12, 2010].
19. England's out-of-hours GP care 'is inadequate'. *BBC.co.uk*. Available at: <http://news.bbc.co.uk/1/hi/health/8565508.stm> [accessed April 12, 2010].
20. Agha R, Cooper D, Muir G. The reporting quality of randomised controlled trials in surgery: a systematic review. *International Journal of Surgery* 2007;**5**(6):413–22.
21. Grumbach K. Chronic illness, comorbidities, and the need for medical generalism. *Annals of Family Medicine* 2003;**1**:4–7.
22. Iona Heath. Dare to use your own intelligence. *British Medical Journal* 2008;**337**:434.
23. Thomas G. The tyranny of the hierarchy of evidence. Cases journal. Comment (4) on Richard Smith's editorial: why do we need cases journal? *Cases Journal* 2008;**1**:1.
24. Siemionow M, Papay F, Alam D, Bernard S, Djohan R, Gordon C, et al. Near-total human face transplantation for a severely disfigured patient in the USA. *The Lancet* 2009;**374**(9685):203–9.
25. Fageeh W, Raffa H, Jabbar H, Marzouki A. Transplantation of the human uterus. *Gynaecology and Obstetrics* 2002;**76**:245–51.
26. Abbasi K. JRSMS short reports – a new online sister journal to JRSMS. *Journal of Royal Society of Medicine* 2010;**103**:1–2.
27. Iain C. Cases journal should follow Venning's quarter-century old example. Comment (4) on Richard Smith's editorial: why do we need cases journal? *Cases Journal* 2008;**1**:1–11.
28. Hill AB. The clinical trial. *New England Journal of Medicine* 1952;**247**:113–9.
29. Rozich JD, Haraden CR, Resar RK. Adverse drug event trigger tool: a practical methodology for measuring medication related harm. *Quality and Safety in Health Care* 2003;**12**:194–200.
30. YellowCard: helping to make medicines safer. Available at: <http://yellowcard.mhra.gov.uk> [accessed on April 15, 2010].
31. National Reporting and Learning Service: practical information, tools and support to improve patient safety in the NHS. Available at: <http://www.nrls.npsa.nhs.uk/home> [accessed April 15, 2010].
32. Lamont T, Luettel D, Scarpello J, O'Driscoll BR, Connew S. Improving the safety of oxygen therapy in hospitals: summary of a safety report from the National Patient Safety Agency. *British Medical Journal* 2010;**340**(January):c187.
33. Britt H, Miller GC, Charles J, Bayram C, Pan Y, Henderson J, et al. *General practice activity in Australia 2006–07*. Canberra: Australian Institute of Health and Welfare; 2008. OpenURL.
34. Surowiecki J. *The wisdom of crowds*. London: Little, Brown; 2004.

Riaz Agha*

Department of Surgery, Cambridge University
Hospitals NHS Foundation Trust,
Cambridge, United Kingdom

R. David Rosin

Academic Surgical Unit, University of the West
Indies, Cave Hill Campus, Bridgetown, Barbados

* Corresponding author.

E-mail addresses: editor@journal-surgery.com
(R. Agha), rdavidrosin@gmail.com (R.D. Rosin)

Available online 4 May 2010