Annals of the
RHEUMATIC DISEASES

Leaders

What value case reports?

"It is certainly a very curious and suggestive case. What do you think Watson?"[1]

Observations based on just one or a few clinical cases have traditionally constituted an acceptable and common format for medical publication. Many specialist journals maintain specific sections for 'case reports' and for aspiring junior doctors this is the usual first arena in which to cut their publishing teeth. However, though case reports can serve several different purposes they are not without their problems (table).

The main justification for case reports is as a principal means of surveillance for rare clinical events. As such they are a common catalyst to the generation of hypotheses concerning pathogenesis, disease associations, management and outcome. Case reports themselves can rarely test the hypotheses they generate but they do place issues before the medical community and often trigger more definitive study. For example, it was largely case reports that suggested numerous metabolic or endocrine diseases as predisposing factors to chondrocalcinosis. Subsequent controlled studies, however, were required to confirm only a minority as true disease associations.[2] A further rationale for case studies is when the complexity or experimental nature of investigations limits their application, logistically or ethically, to just one or a few individuals. Such 'in-depth' reports, although limited in subject numbers, nevertheless can give important insight into mechanisms of disease and treatment.

The principal problem with case reports is that they are very prone to bias. By definition only one or a small number of highly selected patients are examined. These individuals may be unrepresentative of the total patient population – indeed they are inevitably selected because of atypical features. Furthermore, specialist-referred patients are especially targeted. In addition to having a more complex primary condition, such patients are more likely to have multiple pathology. It is recognised, for example, that two conditions which are not associated in the general population can be associated in hospitals if patients with both conditions are referred or admitted at different rates – the 'Berkson' effect.[3] Thus although a report of two conditions coexisting in one or a few patients may be of interest in stimulating hypotheses, the case report itself cannot distinguish chance concurrence from disease association. The reporting of further similar cases may reinforce the rationale for more definitive studies. Importantly, however, such additions to the literature do not answer whether a true association exists. In a large population the issue is not whether rare events occur but whether they occur more frequently than expected by chance.[3] Studies that address prevalence and risk, not further case reports, are required to answer this.

Publishing bias is a further problem. For example, case reports of response to therapy are often misleading since journal editors are more likely to receive and accept reports of dramatic success or disastrous side-effects than less exciting reports of nil or variable response. For both editors and readers the unusual arouses more interest than the mundane. Although often seductive, direct extrapolation from case reports to general experience should be resisted. Only appropriately designed studies can justify and influence routine assessment, management and monitoring.

Because of the many caveats associated with case reports the *Annals* no longer has a section dedicated to their publication. To reach the stage of peer review, submitted case(s) should trigger novel observations, comment or hypotheses with respect to pathogenesis, diagnosis, investigation or management. Repeats of previous observations with no fresh hypothesis or insight ("we report only the fourth such case in the literature") are speedily returned by the Editor. Only a small minority of those submitted are eventually accepted for publication. Successful reports are published principally in the correspondence section as brief communications (limited to 600 words, 10 references, 1 figure and 1 table) with no repetition or undue extrapolation. In depth studies or case(s) of exceptional interest (as judged by the reviewers rather than the authors) may be considered for publication as a concise report.

Case *presentations* are distinct from case *reports*. Reports

The purpose and limitations of case reports

Purpose of report
• surveillance for rare events
• hypothesis generation
• in-depth study of one or a few patients to further understanding of disease mechanism
• sharing experience of unusual clinical experience
Limitations of case reports
• very prone to bias and chance
• cannot be used to determine prevalence, risk or outcome
• usually cannot test hypotheses
• cannot extrapolate to general experience

are aimed at the medical community with the hope of triggering further study. Presentations are directed at the individual reader and when used appropriately serve an important educational role. Though offering no novel or scientific insight, case discussions can expand the limits of clinical experience, reinforce existing competencies and impart new knowledge. Their usefulness as an educational tool reflects the clinician's enthusiasm and interest in clinical stories and the desire to share and discuss experiences with colleagues. A 'cold' review of a clinical topic is inevitably 'warmed up' and made more relevant by focusing the issues and their discussion around an illustrative case. Clinicians readily identify with patients, draw their own conclusions and follow discussion more intently. This is 'reader-centred' rather than 'author-centred' education. The readers are invited to participate (think) as they read. Such a problem-orientated review format is not only more digestible, it also encourages better retention of the teaching points discussed.[4 5] Although complex cases often appear more challenging, important common issues can also be discussed in an interesting way using this format. *"I must admit to you that the case, which seemed to me to be so absurdly simple as to be hardly worth my notice, is rapidly assuming a very different aspect."*[6]

In this issue the 'Masterclass' series returns. In contrast to the previous programme based in Bath, this new series of case presentation and discussion involves different centres in the United Kingdom and mainland Europe. Some clinical messages are worth repeating and from next month's issue we will also publish 'Lesson of the Month' as brief cases with a clear and important practical message. The Editorial Board would welcome submissions for this new case report category. We hope that these two new educational series are read and enjoyed.

Rheumatology Unit,
City Hospital,
Hucknall Road,
Nottingham NG5 1PB,
United Kingdom

MICHAEL DOHERTY

1 Doyle A Conan. The adventure of the creeping man. *The Strand Magazine*, Vol 34, July 1923.
2 Jones A, Chuck A J, Arie E A, Green D J, Doherty M. Diseases associated with calcium pyrophosphate crystal deposition disease. *Sem Arthritis Rheum* 1992; **22:** 188–202.
3 Berkson J. Limitations of the application of fourfold table analysis to hospital data. *Biomed Bull* 1946; **2:** 47–53.
4 Walton H J, Matthews M B. Essentials of problem-based learning. *Medical Education* 1989; **23:** 542–58.
5 Symmons D P M. Continuing medical education for the rheumatologist. *Ann Rheum Dis* 1991; **50:** 453–5.
6 Doyle, A Conan. The Adventure of the Retired Colourman. *The Strand Magazine*, Vol 38, January 1927.