The demise of the published case report – is resuscitation necessary?

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There is general acceptance that discussion of unusual or atypical individual cases can provide an important element in the education of psychiatrists. Case conferences where such patients are presented, are considered an essential requirement of membership training as well as continuing professional development. However, the published form, the case report, has much lower regard and the number of such reports appearing in print has declined substantially over recent years. A brief survey of the *British Journal of Psychiatry* (BJP) covering the years 1987 and 1997 illustrates this trend. Throughout 1987, 44 out of 267 (16%) of articles were case reports compared with only one out of 167 (0.6%) in 1997. Certainly, over the 10-year period, editorial policy for major medical journals has changed, now requiring patients' written agreement prior to the publication of a report of their problems. Although understandable, this adds to the difficulty of publishing reports about patients who are untraceable. However, this is not the only reason for the decline of the published case report. These other reasons have recently been considered by Charlton & Walston (1998) who also argue in favour of the rehabilitation of the case study as a valid method of scientific endeavour. The main points made by the authors will be reviewed and the question of whether the current editorial policy of the BJP should be challenged in light of this will be considered.

CRITICISMS OF CASE STUDY RESEARCH

The main criticisms of case studies are that they are anecdotal, unrepeatable, uncontrolled, unrepresentative and subjectively interpreted. On a more procedural level, case studies are susceptible to misdiagnosis, measurement error, observer misinterpretation or inadequate protocol (Charlton & Walston, 1998). There is also the problem, which occurs not infrequently in psychiatric and other areas of medical research, of too readily interpreting quantitative differences as qualitative. One such example is the clinician who decided to write up the details of a very short patient who had schizophrenia as a case of dwarfism associated with the disorder. The psychiatrist realised his error when he met the patient's parents, both of whom were also quite short. Within the context of evidence-based medicine, case studies represent the lowest form of 'evidence' and are regarded as intrinsically inferior to group studies and large randomised control trials (Sackett *et al*, 1985). A related problem inherent in the case study method is that statistical analysis is not usually possible, which may also have contributed to the view that this is a 'soft' and unscientific approach (Charlton & Walston, 1998).

ROLE OF CASE STUDY RESEARCH IN CLINICAL PSYCHIATRY

Despite these criticisms, there is a long history of case study research in psychology and clinical psychiatry (Tweney *et al*, 1981). The 'ideographic approach' described by Allport (1947) examines the history of the individual and attempts to identify a pattern which is predictive of future behaviour, feelings or attitudes (Hill, 1984). Allport (1961) argued that the examination of groups of subjects (the nomothetic approach) may not always be applicable to the individual since psychological causation is always personal and never actuarial. For example, the risk of a delinquent from a broken home reoffending can be calculated from group frequencies as 80%. However, Allport (1961) makes the point that this percentage has little meaning when applied to the individual who has a 100% certainty of either repeating his crime or of going straight. In other words, group probabilities cannot in actual fact be generalised to the individual (Hill, 1984).

It follows on that while the anecdote cannot be used to derive general principles neither do the rules derived from averaged group observations have meaning at an individual level. The case can therefore be argued that both types of research are necessary. Similarly, in order to progress, scientific endeavour requires occasional changes of direction. This requires investigators to take note of the unexpected event and make new inferences based upon it. This way, new theories can then be derived. All methodologies have limitations and in order to overcome these, several methods, including the case report method, should be employed.

TYPES OF CASE STUDY

Charlton & Walston (1998) suggest that there are two methodological stages in case study research. The first stage is to identify a general theory and then derive from this, specific hypotheses on models which can have implications for the individual case. The second stage is to test these hypothetical models against 'pure' cases which have been selected specifically to exclude confounding variables. The authors go on to describe their two main types of case study. The first of these is the unplanned case observation which challenges existing theory and which the authors term 'serendipity'. The second type, termed 'formal case studies', are those prospectively designed to collect pure cases against which to test a prior hypothesis (Charlton & Walston, 1998). One of the most famous examples of the serendipity process is that of Fleming's observation of the antibacterial effect of penicillin. However, there are many others including the development of several psychopharmacological agents (Healy, 1996). The psychotropic properties of chlorpromazine, iproniazid and imipramine were noted by chance when they were given to treat other disorders. The authors liken their serendipitous process as analogous to that of surveillance for adverse drug effects. Here the alert clinician notes the unexpected: in this case an association between taking a drug and an unusual clinical event. This then leads to the suspicion that the relationship between the two may be causal. In contrast, Charlton & Walston

https://doi.org/10.1192/bjp.174.2.93 Published online by Cambridge University Press

(1998) suggest that formal case studies resemble 'screening' where attempts are made to find specific cases of interest.

An example given by Charlton & Walston concerns theories regarding the psychological causes of persecutory delusions. One such hypothesis proposed by Frith & Corcoran (1996) is that patients develop persecutory delusions because of impaired 'theory of mind'. Charlton & Walston (1998) have been able to demonstrate, using just four carefully selected patients with 'pure' persecutory delusions, that such individuals do perform well on tasks designed to test their 'theory of mind'. Hence, they assert, they have effectively refuted the hypothesis that a defective theory of mind is a necessary factor in the development of persecutory beliefs.

REFUTATIONS AND CONJECTURE

Although Charlton & Walston (1998) have provided a reminder of the importance of single cases or small series, these ideas in science are far from new. Charlton & Walston's implication that their four cases of delusional disorder with intact theory of mind tell us more than would many cases with defective theory of mind, draws upon the philosophical view of Sir Karl Popper. Although somewhat startling when first proposed, Popper's views have now become orthodoxy (Magee, 1973). Popper's theory stated that science properly proceeds by a method of refutation, that is, the correct test of Popper's famous proposal that 'all swans are white' is not to count numerous white individuals but rather to seek out at least one that is black. Case reports are like black swans. For example, a single set of identical triplets with psychosis who were shown to have non-identical psychotic illnesses has been proposed as a refutation of distinct and separate genetic bases for manic–depressive illness and schizophrenia (McGuffin *et al*, 1982). Single cases or small series may also be the starting point for important conjecture, a classic example of which was Alzheimer's description of a single case of the disorder that now bears his name.

More recently, genetic research has provided examples of where the single case or family can yield important clues to the further understanding of psychiatric disorder. Thus, where chromosomal abnormalities are noted to co-occur with mental or other disorders in a single case or family, this can lead to the location of the gene or genes for that disorder. One successful example is that of a patient with a minor deletion on the short arm of the X chromosome which subsequently did much to contribute to the localisation of the Duchenne muscular dystrophy gene (Francke *et al*, 1985). However, even in a comparatively hard area such as genetics one must be prepared for promising conjecture being refuted. An example was the search for a gene for schizophrenia on chromosome 5 (Bassett *et al*, 1988). This followed the observation that a nephew and uncle pair were affected with the disorder and both had a partial trisomy of the chromosome. Many attempts to detect genetic linkage followed but subsequent analysis of all the published data for this region on chromosome 5 has effectively ruled out the location of a gene for schizophrenia at this site (McGuffin *et al*, 1990).

EDITORIAL POLICY

The published case report is still apparently alive and well, although it is more likely to appear in specialised journals than in the BJP. Certainly much scientific research would not have progressed without the insights made on single cases. On the other hand we need to be clear that the usefulness of the case report is limited and it is this author's view that there are only two legitimate reasons for publication. The first is that the case is capable of generating some fresh conjecture that is then amenable to empirical testing. The second is that the case embodies an effective refutation of a particular hypothesis. In the BJP there are clearly fewer such reports appearing now. This may merely reflect that those which have been submitted have not been of a sufficiently high standard rather than because of any deliberate editorial policy against their publication. Charlton & Walston (1998) consider that case study research has been denigrated and neglected and make a worthwhile plea for consciousness-raising on the matter. However, if editors are to encourage authors to consider submitting this type of material to mainstream journals, it must be with the proviso that only those with a high degree of rigour and clarity of purpose will make it into print.

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(First received 24 July 1998, final revision 1 October 1998, accepted 12 October 1998)

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https://doi.org/10.1192/bjp.174.2.93 Published online by Cambridge University Press