

## Role and Evidence of Case Reports and Case Series in Primary Care: A Discussion Paper

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### Abstract

Clinical guidelines, defined as 'systematically developed statements to assist both practitioner and patient decisions in specific circumstances, have become an increasingly familiar part of clinical care. A central challenge in Primary Care is the question of how do we act in the field of atypical courses of diseases or clinical symptoms beside the spectrum of EBM and how could this gap be measured in scientific literature. Main purpose of this paper is a brief analysis of the actual role and evidence of clinical case reports and case series in primary care as a basis for a scientific discussion.

According to the scientific-accepted Oxford levels of evidence case reports and case series belong to the level 4 of evidence. At present a significant majority of published case reports deal with descriptions of physician's care following their clinical intuition beside the pathways of randomized controlled trials (RCT) or guidelines. Clinical symptoms of many cases will add up to what we would recognize as a 'medical' condition, but in up to 19% they are vague, non-specific and/or contradictory and the management of patients with such undefined symptoms can pose a daunting challenge. Even randomized controlled trials are the gold standard in clinical research and systematic reviews and meta-analysis have the highest level of evidence, there is still a need for well-prepared case reports; for instance to describe rare adverse drug effects.

**Keywords:** Case reports; Levels of evidence; Primary care; Clinical intuition; Clinical decision making

### Background

As the name suggests, evidence-based medicine (EBM), is about finding evidence and using that evidence to make clinical decisions. A cornerstone of EBM is the hierarchical system of classifying evidence. This hierarchy is known as the levels of evidence. Physicians are encouraged to find the highest level of evidence to answer clinical questions [1]. According to the scientific-accepted Oxford levels of evidence case reports and case series belong to the level 4 of evidence and as a consequence fulfill the grade C of scientific recommendation in medicine [2]. Clinical guidelines, defined as 'systematically developed statements to assist both practitioner and patient decisions in specific circumstances, have become an increasingly familiar part of clinical care. Guidelines are viewed as useful tools for making care more consistent and efficient and for closing the gap between what clinicians do and what scientific evidence supports [3]. Usually guidelines are based on regular courses of common diseases.

On the other hand a central challenge in Primary Care is the question of how do we act in the field of atypical courses of diseases or clinical symptoms beside the spectrum of EBM and how could this gap be measured in scientific literature.

GP care worldwide is widely associated with a multivariance of clinical cases, which hardly can be detected and covered by scientific guidelines and more and more develops from completely unspecific clinical symptoms to a variety of feared worst case scenarios. Measurement of this multivariance of cases in primary care research often was conducted by using screening tools concerning the reasons for encounter in primary care: Most common reasons for the encounter are musculoskeletal (21.5%) and respiratory (15.2%) symptoms, predominant diagnostic groups, i.e. ICD-10 chapters, are musculoskeletal (17.2%) and respiratory (12.4%). The most common specific diagnoses are essential hypertension (8.1%) and acute upper respiratory infections (3.7%) [4].

### Hypothesis

There is a strong need of case reports in primary care on the best

valuable scientific level. Case reports may assist the decision making process either by providing guidance to general practitioners (GPs) on identifying rarer conditions or a searchable database for looking up seemingly disparate symptoms. One of the main competences of GPs should be a broad spectrum of diagnostic tools in daily practice; therefore case reports reflecting e.g. rare and unexpected courses of a disease may strengthen these competences.

### Objectives

Main purpose of this paper is a brief analysis of the actual role and evidence of clinical case reports and case series in primary care as a basis for a scientific discussion.

#### Four challenges in primary care

#### Unexplained physical symptoms in primary care

Contradictory to clearly defined diagnoses as the most frequent diagnoses encounters for medically unexplained physical symptoms (MUPS) are common in primary health care. Somatization ('experiencing and reporting unexplained somatic symptoms') may indicate concurrent or future disability but this may also partly be caused by psychiatric disorders like anxiety or depressive disorders [5].

Concerning this fact a single cases study was undertaken to assess recognition of medically unexplained physical symptoms by

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general practitioners (GPs), and the feasibility of using a screening procedure based on validated self-report questionnaires. GPs identified unexplained physical symptoms as the main clinical problem for 19% of attending patients. Screening instruments identified 35% of patients as having multiple unexplained physical symptoms, of whom 5% were probable cases of somatization disorder. Nine percent of attending patients reported high levels of health anxiety. Twenty percent were probable cases of mood disorder: in half of these, psychological symptoms were not documented in the case notes. Patients with more somatic symptoms and higher health anxiety were more likely to be recognized by the GP: higher levels of mood symptoms did not predict recognition [6]. According to this level of GP care Robert Braun at first formulated the concept of “preventable dangerous clinical courses in primary care” in the early 1970th [7].

### The GP's role

A central challenge in GP care is the increasing need for prioritization of therapy and treatment decisions mainly within older ages, where the family physician or GP as a local single instance often has to execute significant and vital treatment decisions alone. He should choose appropriate consultants, trying to provide required expertise and compatible personalities to relate with his patient and the patient's family [8,9].

### Clinical intuition in primary care

Studies of clinical intuition, however, equate it to early impressions, the first thing that comes to the physician's mind. A single cases study aimed to investigate the validity of this perspective by examining real cases of intuition in family medicine: 18 family physicians were interviewed about patient cases in which they believed that they had experienced an intuition. Cases were included if (1) participants were unaware of the basis of their judgment, or (2) participants talked about the basis of their judgment but believed that it was irrational or unsubstantiated. In all cases, participants thought that their intuitive judgment was in conflict with a more rational explanation or what other colleagues would do. Automatic, nonanalytical processes in clinical judgment extend beyond first impressions. Rather than admonishing clinicians not to trust their intuition, it should be acknowledged that little is currently known about the different types of intuitive processes and what determines their success or failure [10]. The educational research literature suggests that we can improve our intuitive powers through systematic critical reflection about intuitive judgments-for example, through creative writing (e.g. case reports) and dialogue with professional colleagues [11].

### Diagnostic uncertainty in primary care

In common the prevalence of diseases is low in primary care. Therefore, the positive predictive value of diagnostic tests is lower than in hospitals where patients are highly selected. In addition, the patients present with milder forms of disease; and many diseases might hide behind the initial symptom(s). These facts lead to diagnostic uncertainty which is somewhat inherent to general practice. Fear of uncertainty correlates with higher diagnostic activities. Dealing with uncertainty should be seen as an important core component of general practice [12]. As a direct consequence a scientific dilemma could be observed within the past decades of primary care research: The general practitioner is in a unique position to observe the interaction between the scientific paradigms of evidence based bio-medicine (EBM) and individuals, whether suffering from ill health or considering themselves healthy [13].

## Scientific Rationale of Case Reports and Case Series

A case report is a narrative that describes, for medical, scientific, or educational purposes, a medical problem experienced by one or more patients. Case reports written without guidance from reporting standards are insufficiently rigorous to guide clinical practice, therefore the CARE (CAse REport) guidelines by medical journals will improve the completeness and transparency of published case reports and that the systematic aggregation of information from case reports will inform clinical study design, provide early signals of effectiveness and harms, and improve healthcare delivery [14]. Case reports in primary care research should integrate the following intentions:

- Unexpected and unreported presentations of a disease.
- Management of new and emerging diseases.
- An unexpected and under-reported event in treating a patient.
- A novel and unreported method of management.
- Unreported adverse drug reactions.
- Findings that shed new light on pathogenesis.

Equipment problems.

A short review of the existing literature on an unusual scenario, along with a case report [15].

## The Role of Case Reports and their Impact in Scientific Medical History

At first case reports and single case series presented as ‘letters to the editor’ in the late 1950th and early 1960th emphasized the increasing rate of congenital abnormalities under thalidomide as the scientific basis of further research confirming these early clinical impressions [16,17]. At present a significant majority of published case reports deal with descriptions of physicians care following their clinical intuition beside the pathways of randomized controlled trials (RCT) or guidelines [18-20].

## Case Reports in Primary Care

Unexpected and unreported presentations of a disease often are in the focus of many case reports, e.g. a case report on an amoebiasis-associated colon perforation, presenting the clinical course of 68 years old male patient from Turkey who lived for more than 30 years in Germany and had not been abroad during the past two years. Resistant asymptomatic amoebic dormant bodies caused an emergency-laparoscopy and revealed the seldom complication of a colon perforation. During laparotomy the intraoperative findings mimicked a sarcoma of the mesocolon. The diagnosis could not be assured until the final histopathological results were available. The amoebic colitis was treated with metronidazole followed by paromomycin resulting in successful eradication of the amoebae [21].

Another example-derived from the most frequent reasons for encounter in primary care (neuropathic pain) is a published case series reporting the clinical courses and outcomes in two patients with herpetic neuralgia as a most common reason for encounter, who received a) standard antiviral and analgesic treatment and b) beside the pathways of EBM intravenous ascorbic acid. The cases demonstrated a standard advice and treatment occasion of acute Herpes zoster (HZ) with acute herpetic neuralgia in clinical practice. In these two patients who received intravenous administration of vitamin C as an ad-on therapy a swift regression and clinical improvement of the HZ-

induced rashes, rapid pain reduction and at least a prevention of a later ongoing post herpetic neuralgia could be observed [22]. As a direct consequence the same group of academic GPs therefore conducted a multicenter prospective cohort study concerning the intravenous use of ascorbic acid in shingles providing evidence that concomitant use of intravenously administered ascorbic acid may have beneficial effects on herpes zoster-associated pain, dermatologic findings and accompanying common complaints [23]. Therefore it might be concluded, that he use of drugs outside of approved indications and therapeutic trials can thus inspire to larger trials. Novel and unreported methods of management often are the basis of multiple case reports in primary care research reflecting e.g. pain disorders as the most frequent reasons for encounter and reporting a combination of standardised treatment pathways and use of complementary and alternative medicine (CAM): A clinical case report of a 33-year-old female patient who suffered from a CRPS type I (cold type) of the left foot which had been traumatically induced 9 months before presented the beneficial outcomes of a multidisciplinary therapy-management based on CAM with intensive use of hydrotherapy according to Kneipp, and physiotherapy decreasing significantly pain and local symptoms like hyperhidrosis, trophic skin disorders and lymphedema [18]. Another example is the presentation of the clinical course of a 56-year-old patient, suffering from a severe postradical neck pain syndrome (PRNS) after thyroidectomy and neck dissection due to papillary thyroid cancer since 2 years, who was treated with a combination of hydrotherapy according to Kneipp, conventional physiotherapy, acupuncture and cantharidin patches with a significant reduction of neuropathic pain, increased range of cervical mobility and improvement of health-related quality of life [20].

## Experiences from Other Clinical Disciplines

Concerning the anesthesiology discipline case reports were analyzed with the intention of delineating the trend of case reports over a timespan of 17 years by analyzing the frequency of publication, citation and place of citation: 74.2% of total case reports cited were first cited within 2 years of publication while 34.7% were never cited with the conclusion, that the number of citations of case reports can give us information about the importance of a clinical situation at a particular time [24].

## Discussion

Concerning for instance German ambulatory care physicians' perspectives on clinical guidelines it became obvious that of the total study population 55.3% of physicians reported already using guidelines in the treatment of patients. Physicians in group practices (GrP) as well as general practitioners agreed significantly more with the usefulness of guidelines as a basis for patient care than doctors in single practices or specialists, -33.1% of the participants demonstrated a strong rejection to the application of guidelines in patient care [25]. The intentions of rejection under 1/3 of answering physicians still remain quite unclear: Patient safety was reported to be more important than adherence to guidelines or maintaining a good patient-doctor relationship. Cost containment was perceived both as a motivating factor and a barrier for adherence to guidelines. GPs expressed concerns about difficulties with adherence to guidelines when managing patients with drugs from other prescribers. GPs experienced a lack of time to self-inform and difficulties managing direct-to-consumer drug industry information [26].

In closing this gap case reports may assist the decision making process either by providing guidance to generalists on identifying rarer conditions or a searchable database for looking up seemingly disparate

symptoms. Clinical symptoms of many cases will add up to what we would recognize as a 'medical' condition, but in up to 19% they are vague, non-specific and/or contradictory and the management of patients with such undefined symptoms can pose a daunting challenge. One of the big fears with such patients is that we are missing something, and dealing with undefined symptoms can be unsettling. No generalist can ever expect to know everything about every condition. The research evidence clearly shows that doctors are 'good' at dealing with conditions they treat on a regular basis and potentially over-diagnose those that they have seen recently. One of the key skills in these circumstance is in knowing when and where to look things up [27].

Formulating these intentions we have to keep in mind that it is our privilege and professional duty to reflect upon clinical experience and be open to critical debate; furthermore the publication of case reports and case series should be a scientific basis for critical debates and remarks from the scientific community reflecting potential treatment errors or adding alternative pathways in diagnosis and treatment. However, authors, reviewers, and editors need to ensure that these reports continue to provide new insights into our specialty and that their numbers are controlled. The challenge for editors is to be innovative in turning such material into a product that will enhance the journal's impact [28]. The publication of case reports and case series should be a scientific basis for critical debates and remarks from the scientific community reflecting potential treatment errors or adding alternative pathways in diagnosis and treatment. However, authors, reviewers, and editors need to ensure that these reports continue to provide new insights into our specialty and that their numbers are controlled.

## Conclusion

Clinical decision making can be challenging for both generalists and specialists. Even randomized controlled trials are the gold standard in clinical research and systematic reviews and meta-analysis have the highest level of evidence, there is still a need for well-prepared case reports; for instance to describe rare adverse drug effects.

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