

## Main Article

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# Case report classics in otolaryngology – head and neck surgery: citation analysis

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## Abstract

**Objectives.** To analyse publication and citations trends of case reports within otolaryngology – head and neck surgery literature, with specific attention to the most-cited reports.

**Study design.** Database query.

**Methods.** Web of Science was searched for article type ‘case reports’ published in the leading otolaryngology – head and neck surgery journals since 1945. Variables including publication dates, citation dates and numbers, author, author number, and others were recorded and analysed for trends. The reports with the most citations (classics) were further studied.

**Results.** Of nearly 67 000 published articles in leading otolaryngology – head and neck surgery journals, the overall number of case reports as a percentage of the total has substantially decreased over time. A total of 110 case report classics were identified for which citations have increased.

**Conclusion.** Although the case report may not be worthy of its tarnished record, declining trends in publication suggest a limited future for this valuable research and educational resource.

## Introduction

Case reports are the simplest form of descriptive study, long considered a valuable research and educational resource. They have been the cornerstone of medical education and training for decades. Nonetheless, their publication within the top medical journals has become substantially less commonplace.<sup>1</sup> There are many explanations for this shift, especially in today's era where the importance of evidence-based medicine is stressed. The recent explosion of high-quality research may leave little room for anything else in journals where pages are limited. Moreover, as case reports are relatively low in the hierarchy of clinical decision-making (resting above only expert opinion), publishers may perceive a poor citation rate.<sup>2,3</sup> By publishing only those papers likely to receive a high number of citations, journals may aim to maximally increase their impact factor. The converse, however, is also true. Publishing articles that are poorly cited, as is assumed regarding the case report, has been shown to have a negative effect on the impact factor of a journal.<sup>4</sup>

It remains to be seen whether the case report is worthy of its now tarnished reputation and declining publication rates. This study aimed to analyse trends in the publication and citation of classic case reports within otolaryngology – head and neck surgery, and to determine whether the general perception of low citation rates of case reports within the field is warranted.

## Materials and methods

The Web of Science database (which also included both the Medline and Biosis online databases) was queried on 12 August 2016. Using the filters provided by the database (e.g. ‘document type = case report’, ‘language = English’ and ‘timespan = all years’), the top 10 otolaryngology – head and neck surgery journals (by impact factor, as per our previous study) were searched.<sup>5</sup> Two authors (LWE, DHC) reviewed each article and included only reports that described the medical history of a single patient, as per the standard definition.<sup>6</sup> Reports of multiple cases, including case series, were excluded.

The top 100 most-cited case reports were selected and considered ‘case report classics’. If more than 1 report shared the same number of citations as the 100th most-cited report, they were included. The journals that published the ‘case report classics’ were then queried again for the total number of articles published. Using the same search criteria as above, all reports, irrespective of number of citations, were selected.

## Results

### Case report trends

Between 1945 and 2016, there were 66 953 total articles published by the 10 journals examined in this study. A total of 10 107 were case reports, representing 15.1 per cent

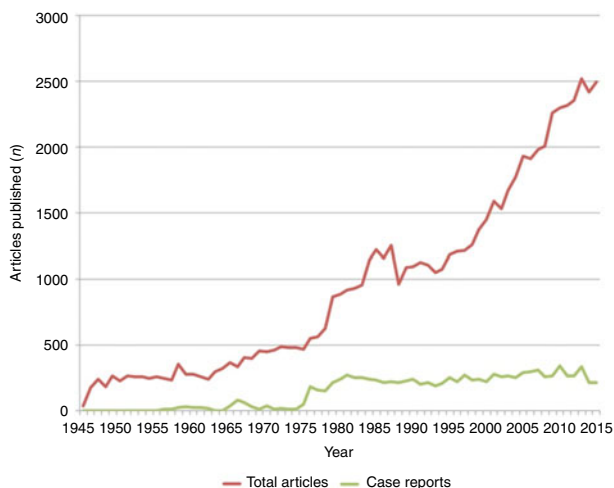


Fig. 1. Case reports and total number of articles published by year.

of all published articles. Over time, the total number of articles published has increased markedly, while the number of case reports has increased marginally (Figure 1). Case reports as a percentage of all published articles decreased steadily over the same time period (Figure 2). By the end of the first decade in the new millennium, there was a consistent downtrend, with only 15.0 per cent of all published articles being case reports. For the most recent years with complete data available, the percentage has continued to decrease, with only 8.82 per cent and 8.5 per cent of all published articles being case reports in 2014 and 2015 respectively.

Case report classics

Between 1945 and 2016, 110 case report classics (as defined above) with 34 or more citations were identified. The average number of citations per case report was 50.5, for a total of 5555 citations. There were 32 (29.1 per cent) case reports with 50 or more citations.

The most-cited case report, ‘A new treatment for chronic secretory otitis media’, by Armstrong, published in *Archives of Otolaryngology – Head & Neck Surgery* (now *JAMA Otolaryngology – Head & Neck Surgery*), had 321 citations, and has been cited an average of 5.1 times per year since publication (Table I). This case report, as well as the case report ‘Unusual neoplasms of the esophagus - review of literature and report of a case’ (by Gregg and Stamler, also published in *Archives of*

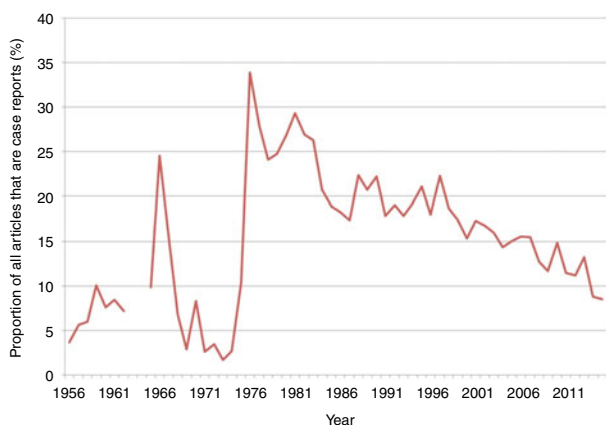


Fig. 2. Case reports as a percentage of total articles published by year.

TABLE I. TOP TEN MOST-CITED OTOLARYNGOLOGY CASE REPORTS

Publication reference details	Citations (n)	Average citations per year (n)
1. Armstrong BW. A new treatment for chronic secretory otitis media. <i>Arch Otolaryngol</i> 1954; <b>59</b> :653–4	321	5.10
2. Montgomery WW. T-tube tracheal stent. <i>Arch Otolaryngol</i> 1965; <b>82</b> :320–1	182	3.50
3. Kraus N, Bradlow AR, Cheatham MM, Cunningham J, King CD, Koch DB et al. Consequences of neural asynchrony: a case of auditory neuropathy. <i>J Assoc Res Otolaryngol</i> 2000; <b>1</b> :33–45	126	7.41
4. Farrugia MC, Summerlin DJ, Krowiak E, Huntley T, Freeman S, Borrowdale R et al. Osteonecrosis of the mandible or maxilla associated with the use of new generation bisphosphonates. <i>Laryngoscope</i> 2006; <b>116</b> :115–20	97	8.82
5. Buckmiller L, Dyamenahalli U, Richter GT. Propranolol for airway hemangiomas: case report of novel treatment. <i>Laryngoscope</i> 2009; <b>119</b> :2051–4	89	11.12
6. Hirshman CA, Smith J. Indirect ignition of the endotracheal tube during carbon dioxide laser surgery. <i>Arch Otolaryngol</i> 1980; <b>106</b> :639–41	83	2.18
7. Veldman JE, Roord JJ, OConnor AF, Shea JJ. Autoimmunity and inner ear disorders: an immune-complex mediated sensorineural hearing loss. <i>Laryngoscope</i> 1984; <b>94</b> :501–7	77	2.26
8. Li JC, Brackmann D, Lo W, Carberry JN, House JW. Reclassification of aggressive adenomatous mastoid neoplasms as endolymphatic sac tumors. <i>Laryngoscope</i> 1993; <b>103</b> :1342–8	76	3.17
9. Rady PL, Schnadig VJ, Weiss RL, Hughes TK, Tying SK. Malignant transformation of recurrent respiratory papillomatosis associated with integrated human papillomavirus type 11 DNA and mutation of p53. <i>Laryngoscope</i> 1998; <b>108</b> :735–40	75	3.95
10. Biller HF, Shugar JM, Krespi YP. A new technique for wide-field exposure of the base of the skull. <i>Arch Otolaryngol</i> 1981; <b>107</b> :698–702	74	2.00

*Otolaryngology*), are the earliest case report classics that met the inclusion criteria, both being published in 1954.

The most recent case report to make the list was ‘Propranolol for airway hemangiomas: case report of novel treatment’ by Buckmiller et al., published in 2009 in *The Laryngoscope*, with 89 citations. This case report also has the highest number of average citations per year since publication, at 11.1, and is within the top 10 most-cited case reports according to our methods (Table I).

Six of the top 10 otolaryngology – head and neck surgery journals (by impact factor) did not publish any classic case reports. The journal *Archives of Otolaryngology* published the most case report classics (n = 52, 47.3 per cent) (Table II). Five out of the top 10 most-cited case report classics were published in *The Laryngoscope*, 4 were in *Archives of Otolaryngology*, and 1 was in *JARO – Journal of the Association for Research in Otolaryngology*. Four of the case

TABLE II. CLASSIC CASE REPORTS BY JOURNAL OF PUBLICATION

Journal title	Classic case reports (n)
Archives of Otolaryngology – Head & Neck Surgery (now JAMA Otolaryngology – Head & Neck Surgery)	52
The Laryngoscope	36
Head & Neck	12
Otolaryngology–Head and Neck Surgery	7
Otology & Neurotology	2
JARO – Journal of the Association for Research in Otolaryngology	1
Total	110

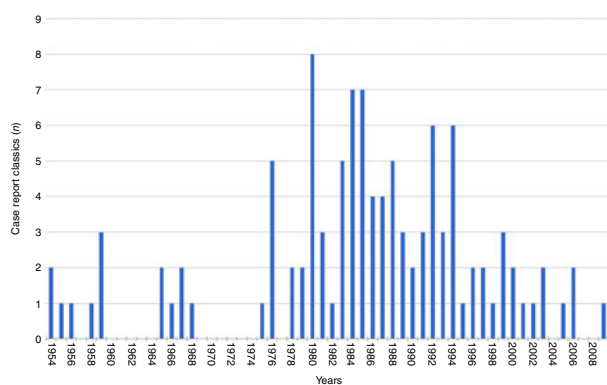


Fig. 3. Case report classics by year of publication.

reports with the highest average per year citation rate were published in *The Laryngoscope*.

The yearly distribution of classic case reports by publication date are displayed in Figure 3. Eight of the case report classics included in this study were published in the 1950s (all published in the journal *Archives of Otolaryngology* (since renamed *JAMA Otolaryngology – Head & Neck Surgery*)). Six

were published in the 1960s and 10 in the 1970s. The bulk of the case report classics ( $n=47$ ) were published in the 1980s. The year with the most case report classics published was 1980, with eight reports. The years 1984 and 1985 each saw the publication of seven classic case reports. The number of classic case reports has steadily declined since then, with 29 published in the 1990s and 10 in the 2000s. No case reports published in the 2010s had enough citations to qualify as a classic. The yearly distribution of the 5555 citations of the 110 case report classics is shown in Figure 4. The year in which the most classic reports were cited was 2008 ( $n=214$ ).

The average number of authors per case report classic is 3.4. The median number of authors is three, where most of the articles had two authors (mode). The case report classic with the most authors is ‘Consequences of neural asynchrony: a case of auditory neuropathy’ by Kraus *et al.* (published in 2000 in *JARO – Journal of the Association for Research in Otolaryngology*), with a total of 10 authors. Only 9 of the 110 case report classics had self-citations. The author with the most case report classics was HF Biller, with five articles. His most-cited case report is ‘A new technique for wide-field exposure of the base of the skull’, published in *Archives of Otolaryngology* in 1981, with 74 citations. That article is also one of the top 10 case reports with the highest average number of citations per year.

Eleven countries were represented in the publication of these case report classics. The majority of the case report classics were written by, or collaborated with, US authors ( $n=102$ , 92.7 per cent), with 99 (90.0 per cent) being strictly US authors. The country with the second most case report classics published was Switzerland, with four articles (3.6 per cent). The UK was represented by authors from three papers, where two of those were collaborations with US authors.

Discussion

Publications in medical journals, especially those with a high impact factor, are viewed as educational currency that demonstrates the level of influence and accomplishment of an author.<sup>7</sup> One measure of the success of a publication is the

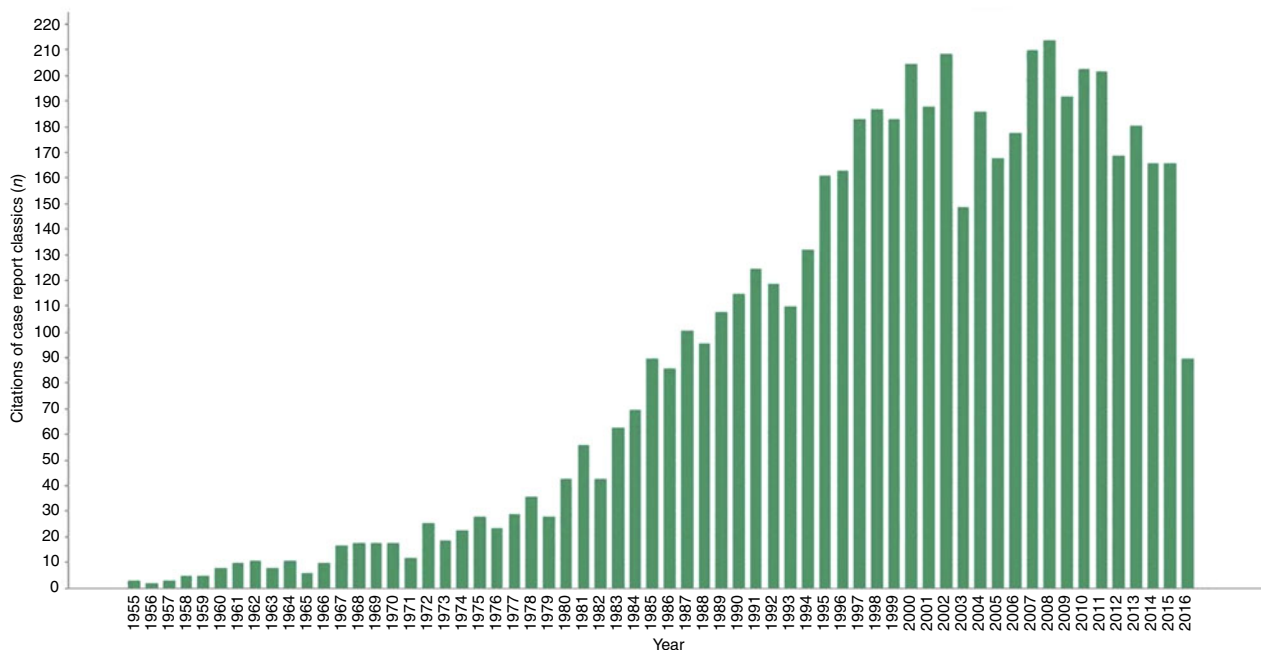


Fig. 4. Citations of case report classics by year.

number of citations it receives, and this number is largely influenced by the strength of the evidence presented in the paper.<sup>8</sup> Citing high levels of evidence provides a more robust background to the results to be presented in each study.<sup>9</sup>

It is often assumed, especially in our current environment of evidence-based medicine, that case reports represent the lowest level of evidence. Many journal editors may consider the case report as ‘filler’ material, as they need little editing and can be inserted as the need arises. Those of the same outlook may similarly presume it to be a platform for novice academics to ‘wet their feet’ and cushion their curriculum vitae.<sup>1</sup> Conversely and pedagogically, we often consider randomised clinical trials, systematic reviews and meta-analyses as the highest level of evidence.<sup>10</sup> Is this the situation the case report has found itself in, that it is mere filler rather an edifying supplemental didactic? Where does the case report fit in with current evidence-based otolaryngology?

In this study, only 3 case reports had over 100 citations and were considered an overall ‘citation classic’, comprising less than 0.33 per cent of the 906 classics found in our previous study.<sup>8</sup> This value acts as either a clarion call for a revival of case reports in otolaryngology – head and neck surgery literature or as verification that there is no demand for case reports in our current condition of publication. The total number of articles published each year in these journals has been increasing, while the proportion of case reports published each year has dramatically decreased since the 1970s. In fact, that number of actual case reports is likely to be substantially lower, as the Web of Science document type definition of ‘case report’ is broader than the ‘single case’ definition.

This decrease may be a result of the position held by some that usually new discoveries from case reports are not supported by additional research, that they may include ‘misleading elements in clinical presentation’, and that they do ‘more harm than good by emphasizing the bizarre’.<sup>11</sup> Yet it is safe to assume that all forms of research have their proper place.<sup>12</sup>

Strict submission criteria enforced by many journals can prevent countless case reports from ever being submitted. Take for instance the journal *Otolaryngology – Head and Neck Surgery*, which requires case reports to be no more than 700 words, does not allow literature reviews, and any case reports describing therapeutic interventions are not considered (as they require higher levels of evidence). Other well-regarded journals, for example *The American Journal of Rhinology & Allergy*, do not accept case reports at all, and refer all case reports to their open access journal. Some journals accept case reports, but also mention in the author guidelines that if it is not accepted it can be published in their open access journal (e.g. *Clinical Otolaryngology*). Such pay-to-publish practices may discourage authors from submitting their work at all. Other high impact factor journals (i.e. *Current Opinion in Otolaryngology & Head and Neck Surgery*, and *Otolaryngologic Clinics of North America*) publish by invitation only and do not publish case reports. Alternatively, there are some journals which still see the value of the case report, such as *Otology & Neurotology* and *JAMA Otolaryngology – Head & Neck Surgery*; these provide space within each journal for a ‘clinical capsule report’, ‘imaging case of the month’ or ‘temporal bone histopathology case of the month’. Other non-open access journals also solicit and accept case reports, but they are in general publications with lower impact factors, and may not be as widely read.

An argument for the case report can be made. The inception of an investigation first begins with an unexpected

observation. Then the observation is documented, and, as paraphrased by Vanderbrouke, becomes ‘refutations of our previously held beliefs...which...leads to new conjectures—new ideas, and new theories’.<sup>11</sup> This then forms into a discussion or a review of many cases. Based on these multiple observations, we can perform a meta-analysis of these case reports and case series, and hopefully reach a significant wake of new conclusions.<sup>13</sup> The educational endeavours ignited by the discussion of these rare events allow for a continual ‘intermingling of surprise, deduction, and induction’ that eventually lead to more formal analyses.<sup>11</sup> Therefore, the highest hierarchical mode of investigation (i.e. randomised control trials) stem from the inception of a question created by a case report. The aim of the evidence-based investigation is ameliorated by the aim of the case report, and the case report rightfully becomes complementary to the modern study.

For example, when BF McCabe began describing cases of patients with autoimmune vasculitides developing sensorineural hearing loss in 1979, more cases of autoimmune disease related to sensorineural hearing loss began appearing (i.e. ulcerative colitis). This illumination led to an increased interest in the pathophysiology, treatment and prevention, and eventually to the greater hierarchical studies performed (randomised control trials), and now to the current (though still incomplete) understanding we have today.<sup>14–16</sup> This was also the case in the retraction of many drugs from the market, prompted by case reports, as well as the discovery of and alternative use for sildenafil as it was being studied as an anti-hypertensive.<sup>1,13,17</sup> In historical medical education, case reports were used to educate on disease processes through individual patient experience of illness and treatment.<sup>18</sup> Case reports catch rare occurrences, add to the disease narrative and advance knowledge of disease pathophysiology that can lead to improved patient care.<sup>7,19</sup> As individual diseases affect each patient differently, it is important to document the often subtle variations of both common and uncommon processes.

The inverse can be argued that the rate of rare occurrences may be decreasing and thus the content of modern case reports is no longer novel. Of interest, the proportion of case reports published each year since the 1970s has been decreasing, whereas the yearly number of citations of case reports has been increasing in the same amount of time. (Note that although Figure 4 reflects a decrease in classic report citations over the last decade, the citations per year to publications per year ratio for case reports is rising.) This inverse relationship may be a sign that the importance of the case report, in the eyes of the journals publishing them, matches its presumed lower level of evidence, resulting in a decrease in overall case report journal acceptance. However, the upward trend noted in case report citation indicates otherwise. Do we as a medical society hold onto the novelties of the past? Has the collective conscious of readers and authors become aware of the lack of case report publications and sought to revive the legacy of those before us? Do we long to recognise Armstrong, Biller, Montgomery and other giants of otolaryngology – head and neck surgery as we peruse and study new findings within the clinical data that are presented in these journals? Is this a sign of a need for increased publication of case reports, or of intrigue in historical medicine?

We can make many assumptions as to why the rare, but highly cited case reports are referenced at the rate that they are. Some introduce a new technique or tool used in disease treatment. Those case reports that outlined treatment procedures remain highly cited. This may be because the results



led to the introduction of tools and treatments that are still used today, such as the grommet, the T-tube tracheal stent, propranolol for airway haemangiomas and the bone-anchored auricular prosthesis. Many of these case reports are cited by publications that further refined and developed treatments and procedures, such as techniques used for wide-field exposure of the skull base. Some case reports are cited by health economists, who examine the cost effectiveness of treatments and evaluate indications for treatment. Others were the first to describe and name a disease process. One case report outlining a disease process of necrotising fasciitis (streptococcal gangrene) of the face, describes its aetiology, pathogenesis, diagnosis and treatment. As such, it is used as the original reference when describing further developments in treatment and pathogenesis. This may be the case where the original article remains the leading citation. It is, however, interesting to think about current otolaryngological diseases that were first introduced as a case report, and yet they are not as highly cited. Could it be these cases and their observed disease state and treatment have been integrated into the collective otolaryngology knowledge base, and thus are not considered to require a citation?

We do not infer that these high impact factor journals have in any way actively tried to quash the existence of the case report in order to protect their well-deserved reputations. It may be that it is becoming increasingly difficult to present a case that has not been presented in some form or fashion already. Whereas the breadth of knowledge continues to grow, these rare occurrences may become rarer.

Woe be unto the reader of a journal who is inundated with multiple case presentations, when the desire for a concentrated form of clinically relevant data is diluted. There is a change in publication trends, whereby journals may publish fewer case reports in favour of publications reporting higher levels of evidence. When so much clinically relevant information is available, why 'waste' space in a journal with a case report, as there are so much clinically relevant data to be presented and discussed within the limited confines of those printed pages.

This could be an argument for a highly impactful otolaryngology – head and neck surgery case report only journal. Yet the formation of such journals (some online), including *BMJ Case Reports – Otolaryngology/ENT*, *Otolaryngology Case Reports*, and *Case Reports in Otolaryngology*, has been achieved but lacks impact. More importantly, the online journals are open access journals that charge a fee for a guaranteed publication. This has arguably turned the publication of case reports into a cacophony of material, and provides limited

- Case reports have research and educational value; however, perceived limited impact has tarnished their reputation
- Publications of case reports within the top otolaryngology – head and neck surgery journals have declined over the last six decades
- Citation rates of the top cited reports continue to increase
- A total of 110 'classic' case reports were identified and analysed for common or differentiating features
- Case reports are poorly represented amongst classic (most-cited) articles in otolaryngology – head and neck surgery
- Although the case report may not be worthy of its tarnished record, a decline in publication suggests a limited future for this valuable resource

merit to its authors or contribution to the otolaryngology – head and neck surgery community.

This study is not without its limitations. While the authors used certain filters and keywords to search for the case reports, we understand that a number of publications may have been overlooked; for instance, true case reports may not have been reported as such within the Web of Science database. We also acknowledge that using only the top 10 most impactful journals excludes possible case reports that may have had a significant number of citations and would have met inclusion criteria otherwise. As the Web of Science database does not have access to articles published before a certain date, this would exclude those articles. One last weakness of the study was the use of a subjective minimum inclusion count of 34 citations. We believe that in order to have a workable dataset, a minimum number of citations needs to be employed. We agreed on setting the minimum number of citations to a number that would bring the total list to approximately 100 case reports.

While the citation rates of case report classics have been evaluated here, future studies could examine submission rates of all case reports, to determine whether their submission rates have fallen in proportion to their publication rates. This is in spite of many previous attempts to revitalise the case report and increase their presence amongst published medical literature.<sup>20</sup>

## Conclusion

Despite the acknowledgement of over 100 'case report classics', the declining presence of case reports amongst published literature may be attributed to an assumed low citation. Multiple influences, including more restrictive submission criteria by journals, the perception of case reports as having a low level of evidence and the theory of decreasing novelty in disease presentations, may keep many case reports from publication. Though the publication of case reports in otolaryngology – head and neck surgery journals is declining, there appears to be an upward trend in the number of citations of case reports. Nevertheless, case reports are very poorly represented amongst citation classics in otolaryngology – head and neck surgery, confirming the conventional belief of low citation rates.

**Competing interests.** None declared

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