The Curious Case of “Case Report” of Infections Caused by Human and Animal Fungal Pathogens: An Educational Tool, an Online Archive, or a Format in Need of Retooling

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Abstract Case reports describe the unusual occurrence and complications of diseases, diagnostic challenges, and notable therapeutic successes. Some journals have discontinued the case reports, while new case report journals have appeared in recent years. During the eightieth anniversary of *Mycopathologia*, it is fitting to examine the relevance of the case report since the journal continues to traverse the boundaries of basic and clinical sciences. A random sample of recent case reports and other articles were selected from *Mycopathologia*. Springer Nature individual article download statistics, and Google Scholar and Scopus citations numbers were compared to assess the reader access and bibliometric impact of case reports. Our analysis indicated that the case report format continues to be a vital element of publication in a cross-disciplinary journal such as *Mycopathologia*. Medical and veterinary case reports covering fungal pathogens are widely read as evident from their download numbers. The download numbers have a positive correlation with the completeness of the

report, the topics and geographic origin of reports have a neutral influence, and the recency leads to lower downloads. There is no discernible trend between the download numbers and the citations of case reports as measured by Google Scholar and Scopus. A specially designed checklist for *Mycopathologia* case reports and new format *Mycopathologia*IMAGES are being introduced to improve the quality and relevance of case reports further.

Introduction

Case reports highlight the unusual disease occurrence and complications, diagnostic challenges, and notable therapeutic successes observed by the astute professionals serving at the frontlines of medical and veterinary practices [1–10]. Case reports have been a regular feature in the most prestigious medical journals including the *New England Journal of Medicine* and *The Lancet*. The ongoing massive changes in the scientific publishing industry have also impacted the publication of case reports [11–16]. Some journals have discontinued the case reports as a feature of their regular contents while new journals, devoted exclusively to case reports, are also being introduced [17–24]. Amidst these unprecedented changes, there is renewed scrutiny of the scholarly impact of the case reports and the case for their continued publication in journals that carry a mixed format of articles [19, 22].

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The case report format is often associated with the variable quality of content, and there is a prevailing view that case reports as a group generate low science citation index measured by the impact factor. However, the overall impact of case reports relative to other publication formats is mixed when an expanded list of benefits and metrics are used for their assessment [25–31]. In a sign of definite progress, a consensus toward standardized format for the case report has emerged among specific disciplines with the use of checklist to ensure high-quality reporting by the prospective authors [32–37].

Mycopathologia had a long history of publishing case reports beginning with the first few inaugural volumes [38–40]. Among these medical and veterinary case reports, there were essential insights into new pathogenic species, new tests, and treatments as well as the undocumented clinical manifestations of well-known pathogenic fungi and fungal diseases. During this eightieth anniversary of publication, we

saw it fitting to re-evaluate the relevance of the case report as a regular publication format in *Mycopathologia* since the journal continues to traverse the boundaries of basic and clinical sciences.

Case Report Analyses

In the following analysis, we focused on the case reports primarily from *Mycopathologia* and their reception in the scientific community. Recent case reports and other articles were selected at random from 2016 to 2017 issues of *Mycopathologia*. Springer Nature individual article download statistics were used to gauge the professional reception of each report after publication (Tables 1, 2, 3, 4, 5, 6, 7; Fig. 1). Google Scholar and Scopus citations numbers of respective reports were compared to the download numbers to assess the bibliometric impact. We also used examples from the *New England Journal of*

Table 1 A sampling of *Mycopathologia* case reports with high downloads (> 200) and high citations (> 3)

Title	Download	Citations ^a	References
First two cases of fungal infections associated with multi-drug resistant yeast, <i>Fereydounia khargensis</i>	1100	3/2	[50]
Subcutaneous phaeohyphomycosis cyst associated with <i>Medicopsis romeroi</i> in an immunocompromised host	998	3/3	[52]
Aspergillosis of the heart and lung and review of published reports on fungal endocarditis	840	6/4	[51]
Fusariosis in a patient with acute myeloid leukemia: a case report and review of the literature	591	10/6	[47]
Fatal cases of bloodstream infection by <i>Fusarium solani</i> and review of published literature	405	5/3	[45]
<i>Paracoccidioides brasiliensis</i> PS2: first autochthonous paracoccidioidomycosis case report in Rio de Janeiro, Brazil, and literature review	334	9/4	[41]
Fungal mycotic aneurysm of the internal carotid artery associated with sphenoid sinusitis in an immunocompromised patient: a case report and review of the literature	325	6/5	[53]
Spondylodiscitis due to <i>Aspergillus terreus</i> in an immunocompetent host: case report and literature review	312	4/2	[43]
Meningitis caused by <i>Candida dubliniensis</i> in a patient with cirrhosis: a case report and review of the literature	297	4/4	[54]
Tinea capitis by <i>Microsporum audouinii</i> : case reports and review of published global literature 2000–2016	284	3/3	[42]
The first case of total dystrophic onychomycosis caused by <i>Aspergillus clavatus</i> resistant to antifungal drugs	255	6/4	[48]
Phaeohyphomycosis due to <i>Exophiala jeanselmei</i> : an emerging pathogen in India-case report and review	209	3/3	[46]
Nebulized liposomal amphotericin B for treatment of pulmonary infection caused by <i>Hormographiella aspergillata</i> : case report and literature review	205	5/2	[44]
Spectral manifestation of melanized fungal infections in kidney transplant recipients: report of six cases	201	4/4	[49]

^aGoogle Scholar/Scopus

Table 2 A sampling of *Mycopathologia* case reports with high downloads (> 200) and low citations (< 2)

Title	Download	Citations ^a	References
Imported talaromycosis in Oman in advanced HIV: a diagnostic challenge outside the endemic areas	820	2/1	[55]
The concept of ecthyma gangrenosum illustrated by a <i>Fusarium oxysporum</i> infection in an immunocompetent individual	794	2/2	[56]
Fournier gangrene caused by <i>Candida albicans</i> in an infant after cardiac surgery	765	1/1	[57]
<i>Rhizopus arrhizus</i> and <i>Fusarium solani</i> concomitant infection in an immunocompromised host	347	2/1	[102]
Primary cutaneous mucormycosis caused by <i>Rhizopus oryzae</i> : a case report and review of literature	302	3/2	[59]
A case report of penile infection caused by fluconazole- and terbinafine-resistant <i>Candida albicans</i>	294	1/1	[60]
The simultaneous occurrence of histoplasmosis and cryptococcal fungemia: a case report and review of the literature	283	0/0	[61]
Urinary tract infection (UTI) caused by <i>Fusarium proliferatum</i> in an agranulocytosis patient and a review of published reports	283	2/0	[64]
Fungal keratitis due to <i>Beauveria bassiana</i> in a contact lenses wearer and review of published reports	268	2/2	[70]
<i>Fusarium dimerum</i> species complex (<i>Fusarium penzigii</i>) keratitis after corneal trauma	260	1/1	[69]
First case of <i>Trichoderma longibrachiatum</i> CIED (Cardiac Implantable Electronic Device)-associated endocarditis in a non-immunocompromised host: biofilm removal and diagnostic problems in the light of the current literature	260	3/1	[66]
<i>Cunninghamella bertholletiae</i> infection in a HLA-haploidentical hematopoietic stem cell transplant recipient with graft failure: case report and review of the literature	220	1/1	[65]
<i>Pseudallescheria boydii</i> with <i>Aspergillus fumigatus</i> and <i>Aspergillus terreus</i> in a critically ill hematopoietic stem cell recipient with ARDS	210	2/2	[68]
Respiratory tract infection caused by <i>Fonsecaea monophora</i> after kidney transplantation	205	1/0	[62]
Combined orofacial aspergillosis and mucormycosis: fatal complication of a recurrent paediatric glioma-case report and review of literature	201	1/0	[67]
Severe paracoccidioidomycosis in a 14-year-old boy	200	1/1	[63]

^aGoogle Scholar/Scopus**Table 3** A sampling of *Mycopathologia* case reports with low downloads (< 200) and high citations (> 3)

Title	Download	Citations ^a	References
Dermatophytosis due to <i>Microsporum incurvatum</i> : Notification and identification of a neglected pathogenic species	199	7/7	[71]
The first case of invasive mixed-mold infections due to <i>Emericella nidulans</i> var. <i>echinulata</i> and <i>Rasamsonia piperina</i> in a patient with chronic granulomatous disease	195	4/4	[72]
Onychomycosis associated with <i>Exophiala oligosperma</i> in Taiwan	165	5/3	[73]
Paracoccidioidomycosis mimicking sarcoidosis: a review of 8 cases	164	5/3	[74]

^aGoogle Scholar/Scopus

Medicine to strengthen our conclusions about the bibliographic measurements and impact of the case reports (Table 8).

High Downloads and Citations

Eight of the 11 case reports in the sample set were downloaded more than 300 times each (Table 1). These case reports were also cited 3 or more times in

Table 4 A sampling of *Mycopathologia* case reports with low downloads (< 200) and low citations (> 2)

Title	Download	Citations ^a	References
<i>Ustilago echinata</i> : infection in a mixed martial artist following an open fracture	169	1/1	[85]
Phaeohyphomycosis caused by <i>Rhytidhysterium rufulum</i> and review of literature	167	3/0	[78]
<i>Lichtheimia</i> infection in a lymphoma patient: case report and a brief review of the available diagnostic tools	157	1/1	[82]
Coccidioidomycosis in patients with selected solid organ cancers: a case series and review of medical literature	154	1/1	[84]
Successful antifungal combination therapy and surgical approach for <i>Aspergillus fumigatus</i> suppurative thyroiditis associated with thyrotoxicosis and review of published reports	146	1/0	[80]
Coinfection with <i>Lichtheimia corymbifera</i> and <i>Aspergillus flavus</i> in an immune-competent patient mimicking as pulmonary-renal syndrome	126	1/1	[81]

^aGoogle Scholar/Scopus**Table 5** A sampling of *Mycopathologia* case reports with low downloads (< 200) and no citations

Title	Download	Citations ^a	References
Acute disseminated <i>Talaromyces marneffei</i> in an immunocompetent patient	180	0/0	[77]
Fungal keratitis secondary to <i>Trametes betulina</i> : a case report and review of literature	162	0/0	[86]
A 9-month-old girl from Iran with extensive erythematous plaques due to <i>Trichophyton simii</i> , a zoophilic dermatophyte	160	0/0	[79]
Gastroesophageal reflux disease and pulmonary diseases associated with aspergillosis: Is there a connection?	148	0/0	[83]
Invasive <i>Aspergillus</i> laryngotracheobronchitis in an adult with primary CNS lymphoma	139	0/0	[87]
Molecular identification of cutaneous alternariosis in a renal transplant patient	111	0/0	[75]
Acute paracoccidioidomycosis in a 40-year-old man: a case report and literature review	107	0/0	[76]
Nosocomial rhinocerebral mucormycosis: two cases with a temporal relationship	95	0/0	[103]

^aGoogle Scholar/Scopus

the last 2 years. These case reports contained a balanced account of case history, mycology and pathology investigations, treatment details, and the review of the relevant literature. They originated from Brazil [41, 42], France [43, 44], India [45, 46], Italy [47], Iran [48], Japan [49], Malaysia [50], Poland [51], UK [52], and USA [53, 54]. There were no other discernible features among this group of case reports to account for the high downloads and citations except for a novelty or rarity of fungal pathogens, for example reports of two cases of multidrug-resistant yeast (*Fereydownia khargensis*) and of black-grain eumycetoma caused by the rare fungal pathogen *Medicopsis romeroi* [50, 52]. Next, we examined the case reports that had either high downloads and low citations, or low downloads and high citations (Tables 2, 3). Unlike the pattern in Table 1, many case reports with

downloads more than 300 were cited once or twice [55–59]. None of the reports included any unusual pathogens, nor there were any multidrug-resistant pathogens reported except for a case of fluconazole- and terbinafine-resistant *Candida albicans* [60]. The geographic origin of these case reports were quite widespread: four each originated from Brazil [58, 61–63] and China [56, 60, 64, 65], two from Italy [66, 67], and one each from Germany [68], Mexico [59], Oman [55], Poland [57], Portugal [69], and Spain [70]. Unlike high download–low citation reports, there are only a few case reports with low downloads and high citations (Table 3). These reports deal with rare pathogens, mixed infection, or unusual clinical presentation in many cases [71–74]. Seventeen of twenty-four reports appeared in 2016, while only seven were published in 2017, suggesting that a

Table 6 A sampling of *Mycopathologia* veterinary case reports

Title	Downloads	Citations ^a	References
Coccidioidomycosis in biopsies with presumptive diagnosis of malignancy in dogs: report of three cases and comparative discussion of published reports	1000	1/1	[88]
Disseminated candidiasis in a young, previously healthy, dog and review of literature	800	1/1	[104]
<i>Cryptococcus neoformans</i> var. <i>grubii</i> -induced arthritis with encephalitic dissemination in a dog and review of published literature	259	8/8	[90]
Detection of multiple budding yeast cells and a partial sequence of 43-kDa glycoprotein coding gene of <i>Paracoccidioides brasiliensis</i> from a case of lacaziosis in a female pacific white-sided dolphin (<i>Lagenorhynchus obliquidens</i>)	245	7/6	[94]
<i>Paracoccidioides brasiliensis</i> -associated dermatitis and lymphadenitis in a dog	224	6/5	[91]
Evidence of a <i>Prototheca zopfii</i> genotype 2 disseminated infection in a dog with cutaneous lesions	197	1/1	[92]
Intestinal lesion in a dog due to <i>Cryptococcus gattii</i> type VGII and review of published cases of canine gastrointestinal cryptococcosis	177	3/4	[105]

^aGoogle Scholar/Scopus**Table 7** A sampling of *Mycopathologia* articles covering clinical and experimental mycology, and opinions

Title	Download	Citations ^a	References
<i>Candida albicans</i> / <i>Staphylococcus aureus</i> dual-species biofilm as a target for the combination of essential oils and fluconazole or mupirocin	1300	2/1	[101]
Challenges in <i>Fusarium</i> , a trans-kingdom pathogen	1100	12/11	[100]
Pathogenic dermatophytes survive in nail lesions during oral terbinafine treatment for Tinea unguium	734	0/0	[99]
Has <i>Aspergillus</i> PCR come to the age of maturity?	714	1/1	[95]
Genotyping of <i>Fusarium</i> isolates from onychomycoses in Colombia: detection of two new species within the <i>Fusarium solani</i> species complex and in vitro antifungal susceptibility testing	453	12/9	[98]
The AAA ATPase Vps4 plays important roles in <i>Candida albicans</i> hyphal formation and is inhibited by DBE4	398	3/0	[96]
Quantitative real-time PCR and Platelia galactomannan assay for the diagnosis of invasive pulmonary aspergillosis: Bronchoalveolar lavage fluid performs better than serum in non-neutropaenic patients	296	9/9	[97]

^aGoogle Scholar/Scopus

longer duration from the date of publication might have some influence both on downloads and on citations. In summary, the case reports that described rare or novel pathogens, multidrug resistance, or unique clinical presentations, appeared in 2016 than in 2017, were downloaded more often and received higher citations. However, a high download did not predict higher citation or vice versa.

Low Downloads and Citations

Next, we examined whether there was any discernible pattern for the case reports that had low downloads and low or no citations (Tables 4, 5). These case reports were reported from Belgium [75], Brazil [76], China [77], India [78], Iran [79], Italy [80], Pakistan [81], Spain [76], Switzerland [82], UK [83], and USA [84–87]. The cases included four *Aspergillus* infections, three pathogens belonging to *Mucorales* order, two basidiomycetes, one each *Alternaria* species,

Table 8 A limited sampling of case reports from the *New England Journal of Medicine*

Title	Diagnosis	Page views	Citations ^a	References
Case records of the Massachussets General Hospital. Case 5-2016. A 43-year-old man with altered mental status and a history of alcohol use	Cerebral mucormycosis	24,931	3/1	[106]
Case 6-2017. A 57-year-old woman with fatigue, sweats, weight loss, headache, and skin lesions	IgG4-related disease	24,475	2/0	[107]
Case 6-2018: A 35-year-old woman with headache, subjective fever, and anemia	Transient aplastic crisis due to parvovirus B19 infection	24,452	2/0	[108]
Case records of the Massachussets General Hospital. Case 4-2016. A 58-year-old woman with a skin ulcer, fever, and lymphadenopathy	Ulceroglandular tularemia	22,473	1/1	[109]
Case records of the Massachussets General Hospital. Case 6-2016. A 10-year-old boy with abdominal cramping and fevers	Adenocarcinoma of colon	20,200	1/4	[110]
Case 5-2017. A 19-year-old man with hematuria and a retroperitoneal mass	Soft tissue sarcoma	19,730	3/2	[111]
Case 5-2018: A 63-year-old man with confusion after stem cell transplantation	Human herpesvirus 6-related encephalitis	15,621	0/0	[112]
Case 4-2018: A newborn with thrombocytopenia, cataracts, and hepatosplenomegaly	Congenital rubella syndrome	12,861	1/1	[113]
Case 4-2017. A 2-month-old girl with growth retardation and respiratory failure	X-linked periventricular heterotopia caused by an FLNA mutation	11,520	0/0	[114]

^aGoogle Scholar/Scopus

Case Reports in New England Journal of Medicine

Among the most acclaimed scientific publications, the *New England Journal of Medicine* includes one case report for almost every week of its yearly publication. We did a random sampling of nine reports for the last 3 years reported in the same month (Table 8). The authors of these case presentations are from the Boston area, and affiliated with the local medical institutions. As expected, these reports were downloaded many thousands of times. Interestingly, recency and topics had no influence on the rates of downloads with a mucormycosis case being most downloaded in this small set. Strikingly, tens of thousands of downloads later, there were still very few citations of these cases. These observations suggest that case reports continue to have an educational role, and it will be a fallacy to subject evaluation of their impact on a single metric such as citation number.

Checklists

As mentioned in the opening section, many journals have instituted a checklist for quality control of case reports being submitted for publication. The essential elements of writing a case report with the help of the checklist are covered in detail elsewhere [32–36]. After examining variable quality of the case reports being submitted to *Mycopathologia*, we propose to introduce a checklist to help the prospective authors with the preparation and publication of high-quality case reports (Table 9). We also observed that not all case reports need the length and description of a typical article to convey their important messages. Therefore, we are introducing *Mycopathologia*IMAGES as a new feature in the journal for the communication of brief clinical observations of interests to the physicians and mycologists (Table 10).

Table 9 A checklist for submission of case report to *Mycopathologia*

	Yes	No
Original observation	☐	☐
Ethical statement included	☐	☐
Patient demographic anonymized	☐	☐
Title includes the term for medical and veterinary condition being described and the correct name of the fungal pathogen (italicized)	☐	☐
Title and abstract do not include superlatives such as ‘new’, ‘first’, ‘unique’, etc.	☐	☐
Patient treatment and follow-up included	☐	☐
Confirmatory, probable or possible diagnostic criteria defined	☐	☐
<i>Mycology</i>		
Any author a specialist in fungal diagnostics?	☐	☐
Culture positive with characteristic microscopy	☐	☐
Culture confirmed with molecular tests	☐	☐
Fungus name conform to current nomenclature	☐	☐
Nucleotide sequence deposited in a known database	☐	☐
Antifungal susceptibility test results included	☐	☐
Antifungal test result interpretation provided with reference to the guidance documents (CLSI, EUCAST)		
<i>Histopathology</i>		
Any author a specialist in histopathology?	☐	☐
High-resolution images included	☐	☐
Specialized fungal stains used	☐	☐
Differential histopathology diagnosis discussed	☐	☐

Table 10 *Mycopathologia* IMAGES checklist

Original, unpublished observation being reported with ethical statement	☐
Report of potential interest to a mixed audience of physicians and laboratory scientists	☐
Brief title (three-four keywords)	☐
One paragraph description not to exceed 500 words	☐
No abstract, literature citation, bibliography or hyperlinks	☐
Maximum of four images with short legends (colored required for free online publication)	☐
Author and primary affiliation at the end of the paragraph (name, degree, primary institution name, city, state, country)	☐

Conclusions

Case reports constitute an essential element of publication in a cross-disciplinary journal such as *Mycopathologia*. Medical and veterinary case reports covering fungal pathogens are widely read as evident from their download numbers. The completeness of the case report appears to have a positive effect on the downloads; the topics and geographic origin have a neutral influence; and recency shows a negative

correlation. There is no correlation between downloads and the citations of case reports as measured by Google Scholar and Scopus. The use of the checklists for the authors and a new format termed *Mycopathologia* IMAGES are likely to improve the quality and relevance of case reports.

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