Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

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TITLE

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

AUTHOR

Serina Stretton, PhD CMPP

AUTHOR'S AFFILIATION

ProScribe Medical Communications, Noosaville, Queensland, Australia

AUTHOR FOR CORRESPONDENCE

Serina Stretton, PhD, CMPP

ProScribe Medical Communications

18 Poulet St

Matraville, NSW, 2036

AUSTRALIA

Tel: + 612 9661 0768

Fax: + 612 9661 0766

Email: ss@proscribe.com.au

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ABSTRACT

Background: Ghostwriting of industry-sponsored articles is unethical and is perceived to be common practice in the medical literature.

Objective: The conduct a systematic review on how evidence for the prevalence of ghostwriting is reported in the medical literature.

Data sources: MEDLINE via PubMed 1966+, EMBASE 1966+, The Cochrane Library 1988+, Medical Writing 1998+, The AMWA Journal 1986+, Council of Science Editors Annual Meetings 2007+, and the Peer Review Congress 1994+ were searched electronically (23 May 2013) using the search terms ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*.

Eligibility criteria: All publication types were considered; only publications reporting a numerical estimate of possible ghostwriting prevalence were included.

Data extraction: Two independent reviewers screened the publications; discrepancies were resolved by consensus. Data to be collected included a numerical estimate of the prevalence of possible ghostwriting (primary outcome measure), definitions of ghostwriting reported, source of the reported prevalence, publication type and year, study design, and sample population.

Results: Of the 848 publications retrieved and screened for eligibility, 48 reported numerical estimates for the prevalence of possible ghostwriting and were analyzed further. Sixteen primary publications reported findings from cross-sectional surveys or descriptive analyses of published articles and 32 secondary publications cited published or unpublished evidence. Estimates on the prevalence of possible ghostwriting in primary and secondary publications varied markedly. Estimates were influenced by the definition of ghostwriting used, the type of population or sample assessed, and whether evidence from primary publications was cited correctly or appropriately.

Conclusions: Estimates of ghostwriting reported in the medical literature can be outdated, misleading, or mistaken. Researchers should not inflate estimates using nonstandard definitions

of ghostwriting nor conflate ghostwriting with other unethical authorship practices. Editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

MAIN STRENGTHS AND LIMITATIONS

Strengths

- First systematic review on the reporting of the prevalence of ghostwriting in the medical literature
- Abroad search strategy was employed with few restrictions to minimize any potential for publication or language bias; there were no restrictions on language.
- All study designs and publication types were considered

Limitations

- Study heterogeneity in the outcomes reported and populations investigated precluded synthesis of the data
- Retrospective and self-reported nature of the data collection resulted increased the risk of selection bias in the studies
- Most included studies were not broadly representative of the peer-reviewed medical literature

INTRODUCTION

Ghostwriting of peer-reviewed journal publications in the medical literature is believed to be common practice.[1-3] This belief is supported, to a large extent, by highly publicized cases, primarily from the 1990s and early 2000s, of pharmaceutical companies and authors who had used ghostwriters to prepare manuscripts for publication in medical journals.[4-6] Such cases are highly unethical because the role of the commercial sponsor and any other potential conflicts of interest were hidden.

Ghostwriting occurs when writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments. This practice is distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[7] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding the two unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[8] Estimating the prevalence of ghostwriting has also been hindered by the failure of authors to distinguish ghostwriting, which is unethical, from professional medical writing support, which is ethical.[9, 10] Ghostwriters keep their involvement in a manuscript hidden, whereas professional medical writers disclose their involvement and follow ethical publication practices.[7, 9-11] The prevalence of disclosed professional medical writing assistance in medical journals has been estimated to be between 6.0% and 11.0%[10, 12] and the legitimate value that this medical writing assistance can bring to improving the quality, timeliness, and integrity of reporting in medical journals has been demonstrated.[7, 13, 14] However, the exact prevalence of

ghostwriting and other forms of undisclosed contributions to papers published in medical journals is unknown.

The primary objective of this study was to conduct the first systematic review on how the prevalence of ghostwriting is reported in the medical literature. The secondary objectives were to assess the variability of the reported estimates of the prevalence of ghostwriting and investigate the source for these estimates.

MATERIALS AND METHODS

Literature search strategy

A search strategy was developed to retrieve publications from the medical literature that reported quantitative estimates of the prevalence of ghostwriting. The following databases were searched on 23 May 2013: MEDLINE via PubMed (1966+); EMBASE (1966+); The Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials (1988+); The AMWA Journal (1986+, journal of the American Medical Writers Association); Medical Writing (1998+, journal of the European Medical Writers Association, formerly known as The Write Stuff); Council of Science Editors annual meetings (2007+); and the International Congress on Peer Review and Biomedical Publication meetings (1994+). General text was searched using the following search terms: ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*. Truncation symbols and Boolean operators (AND, OR) were used wherever possible.

Two reviewers (SS, SMG) independently screened the title and abstracts of all retrieved publications using prespecified eligibility criteria; the full text of potentially eligible publications

was screened to confirm eligibility for inclusion. Any discrepancies between the reviewers were resolved by consensus. The reference lists of relevant reviews and other publications were screened by hand to identify any additional publications for inclusion.

Eligibility criteria

Publications were included if they reported a numerical estimate of the prevalence of possible ghostwriting. Publications were excluded if they were duplicate publications from different databases or abstracts of subsequently published full-text articles, did not report any outcomes related to ghostwriting, or reported qualitative estimate(s) of the prevalence of possible ghostwriting. No restrictions on language were included in the eligibility criteria.

Data extraction and analysis

Data to be collected were prespecified and included publication type and year, study design and sample population, definitions of ghostwriting reported, reported prevalence of possible ghostwriting, and the source of the reported prevalence of ghostwriting.

The primary outcome measure for the systematic review was a numerical estimate of the prevalence of possible ghostwriting. The reported definitions of possible ghostwriting were categorized according to the following standard definitions, irrespective of the term used to describe the practice in the publication. Undisclosed writing contributions to a manuscript were defined as (i) ghostwriting if they were described as not meriting authorship and were not listed in the acknowledgments[9, 15] and (ii) ghost authoring if they did merit authorship and were not listed in the author byline.[7] Consistent with recommendations from international medical journal editors,[13, 16] Good Publication Practice guidelines,[11] and professional medical writing associations,[17, 18] disclosed writing contributions to a manuscript that did not merit authorship and were disclosed in the acknowledgments were not categorized as ghostwriting.

RESULTS

Publication selection

A total of 848 articles were retrieved from the literature search, 800 were excluded, and 48 met the eligibility criteria and were included in the systematic review (Figure 1). The main reasons for exclusion were publications not relevant to ghostwriting (n = 539), duplicate publications from different databases (n = 129), and publications not reporting a numerical estimate of the prevalence of possible ghostwriting (n = 124). The titles or abstracts of the 8 publications that were excluded because the full text or abstract could not be retrieved were reviewed; none were considered to report a numerical estimate of the prevalence of possible ghostwriting. Overall, eligible publications included 16 primary publications that reported original research on the prevalence of possible ghostwriting and 32 secondary publications that cited published or unpublished evidence of possible ghostwriting.

Primary publications

Publication characteristics

Of the 16 primary publications (Table 1), there were 13 full-text publications[4, 6, 19-29] and three conference abstracts[30-32] that reported a numerical estimate of possible ghostwriting. Twelve publications[19-27, 30-32] reported findings from cross-sectional surveys and four publications[4, 6, 28, 29] reported findings from descriptive analyses of published articles. While there is no consensus on the best practice for reporting survey research,[33] most cross-sectional surveys were reasonably well reported. The surveys used were not validated but most included pretested questions, required an anonymous response, were conducted in targeted populations (ie, all individuals invited to participate were involved in the preparation of peer-reviewed manuscripts), and reported a sample size and response rate. Of the cross-sectional surveys conducted in targeted populations (eg, corresponding or first authors), six had response

rates greater than 50%, two had response rates less than 50%, and one did not report a response rate (Table 1).

Of the cross-sectional surveys conducted in nontargeted populations (eg, individuals invited to participate who may or may not have been involved in providing medical writing assistance for peer-reviewed publications), three had low (12% to 28%) response rates and one did not report a response rate (Table 1).

Table 1. Characteristics of Primary Publications Reporting Original Evidence of Ghostwriting

Publication	Population	Survey description	Data collected	N	RR
Cross-sectional surveys – authors or corresponding authors					
Flanagin 1998[26]	6 general medicine journals [a]	Self-administered, postal, anonymous, pretested, targeted	1996	809	69%
Mowatt 2002[22]	Published Cochrane reviews	Self-administered, online, anonymous, pretested, targeted	1999	362	63%
Hao 2009[31] (Abstract)	Chin Med J	Self-administered, email, not anonymous, targeted	2008 [c]	220	86%
Dotson 2011[27]	3 pharmacy journals	Self-administered, online, anonymous, pretested, targeted	2009	112	25%
Mirzazadeh 2011[23]	3 Iranian journals	Self-administered, email, not anonymous, targeted	2009-2010	NR	NR
Wislar 2011[19]	6 general medicine journals [b]	Self-administered, online, anonymous, pretested, targeted	2008	622	70%
Ghajarzadeh 2012[25]	Arch Iran Med (based on student theses)	Self-administered, email, anonymous, targeted	2005-2007	30	49%

Publication	Population	Survey description	Data collected	N	RR
Vinther 2012[20]	Ugeskr Laeger & Dan Med J	Self-administered, online, anonymous, pretested, targeted	2010	272	62%
Cross-sectional surveys – healthcare professionals					
Price 2000[21]	Health academic staff	Self-administered, postal, anonymous, pretested, targeted	NR	166	59%
Rees 2013[30] (Abstract)	Registered users of EPG Online	Self-administered, online, anonymous, nontargeted	NR	295	NR
Cross-sectional surveys – members of medical writing associations					
Jacobs 2009[24]	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2005 2008	843 773	28% 14%
Hamilton 2012[32] (Abstract)	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2011	620	12% [d]
Publication reviews and descriptive analysis					
Healy 2003[4]	Articles on sertraline	NA	1998-2000	96	NA
Gøtzsche 2007[29]	Articles on Danish industry-initiated trials approved 1994-1995	NA	NR	44	NA
Ross 2008[6]	Reviews on rofecoxib associated with Merck support	NA	1996-2004	72	NA
Suda 2011[28]	Noninferiority clinical trials	NA	1989-2009	583	NA

[a] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, American Journal of Cardiology, American Journal of Medicine, American Journal of Obstetrics and Gynecology.

[b] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, Lancet, Nature Medicine, PLoS Medicine.

[c] Submission date.

[d] Personal communication, C. Hamilton.

Abbreviations: AMWA, American Medical Writers Association; EMWA, European Medical Writers Association; NA, not applicable; NR, not reported; RR, response rate.

Not all cross-sectional surveys were broadly representative of the peer-reviewed medical literature. The cross-sectional surveys were conducted in single populations of academic staff or healthcare professionals[21, 30], members of medical writing associations[24, 32], or corresponding or first authors. The corresponding or first authors were surveyed from single journals (Cochrane reviews, Chinese Medical Journal, Archives of Iranian Medicine),[22, 25, 31] two Danish journals,[20] three Iranian journals,[23] and three pharmacy journals.[27] Two cross-sectional surveys of corresponding authors were conducted in six general medicine journals.[19, 26]

The descriptive analyses of published articles were conducted in single populations and included an analysis of sertraline publications from 1998 to 2000,[4] rofecoxib reviews from 1996 to 2004,[6] publications from industry-initiated trials in Denmark from 1994 to 1995,[29] and from published noninferiority trials from 1989 to 2009.[28]

Reported prevalence of ghostwriting

The reported prevalence of possible ghostwriting in the primary publications varied markedly and was difficult to compare because of the different populations assessed, methods used to generate the estimates, and definitions that were used (Tables 1 and 2). All cross-sectional surveys reported a definition of ghostwriting or ghost authoring, but most definitions did not differentiate contributions that merited authorship from those that did not merit authorship (Table 2).

Table 2. Primary Publications Reporting Estimates of the Prevalence of Possible Ghostwriting

Publication	Reported measure	Estimate % (n)
Cross-sectional surveys reporting ghostwriting		
Flanagin 1998[26]	Unnamed individual who participated in the writing	1.4% (11/809) of articles
Wislar 2011[19]	Unnamed individual who participated in the writing	0.2% [a] (1/622) of articles
Jacobs 2009, Hamilton 2012[24, 32]	Undisclosed medical writing assistance not qualifying for authorship	2005: 61.8% (NR) 2009: 41.7% (NR) 2011: 33.0% (NR) of writers
Cross-sectional surveys reporting combined ghost authoring and ghostwriting		
Flanagin 1998[26]	Failure to name, as an author, individuals who made substantial contributions to the research or writing or an unidentified medical writer	11.5% (93/809) of articles
Price 2000[21]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	24.1% (40/166) of authors
Mowatt 2002[22]	Individual merited authorship or had assisted with drafting but not listed as an author or acknowledged	8.8% (32/362) of articles

Publication	Reported measure	Estimate % (n)
Hao 2009[31]	English-language speakers assisted with writing but not identified as authors or acknowledged	10.4% (NR) of authors
Dotson 2011[27]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	0.9% (1/112) of articles
Wislar 2011[19]	Failure to name, as an author, individuals who made substantial contributions to the research or writing of the article or an unnamed individual who participated in the writing	7.9% (49/622) of articles
Cross-sectional surveys reporting ghost authoring		
Mirzazadeh 2011[23]	Failure to name, as an author, individuals who made substantial contributions to the research	21.4% (25/NR) of authors
Ghajarzadeh 2012[25]	Failure to name, as an author, students who made substantial contributions to the research[b]	0.7% (2/296) of articles
Vinther 2012[20]	Individual merited authorship but not listed as an author	2.4% (6/245) of articles
Rees 2013[30]	Individual merited authorship but not listed as an author	70% (NR/202) of published authors
Publication reviews and descriptive analysis reporting possible ghost authoring or ghostwriting		
Healy 2003[4]	Published articles coordinated by a medical information company, including acknowledged medical writing support[c]	57.3% (55/96) of articles
Gøtzsche 2007[29]	Individuals who wrote the trial protocol, conducted the statistical analyses, or wrote the manuscript but were not listed as authors, not members of a study group or steering committee, or not disclosed in an acknowledgment	75.0% (33/44) of trials
Ross 2008[6]	Published reviews associated with Merck support and with a single external author[d]	69.4% (50/72) of reviews

[a] Available as online supplementary data.

[b] Students were classified as ghostwriters if the student was not named as an author and if the results reported in the publications were based on the results of their theses.

[c] Authors conclude data provide quantification of the possible extent of ghostwriting based on a single drug. Of the 55 published articles that were coordinated through a medical information company, 2 included medical writing assistance that was acknowledged in the published article.

[d] Published review articles had been identified from correspondence between Merck and a medical publishing company, from Merck publication status reports, or were affiliated with an author named within the correspondence or publication status reports. The authors did not report whether medical writing assistance was acknowledged in the published article.

Abbreviations: NR, not reported.

Four cross-sectional surveys reported the prevalence of ghostwriting where the definition provided could be categorized as undisclosed contributions that do not merit authorship.[19, 24, 26, 32] Findings from these surveys, which were repeated at different time points, suggested that the prevalence of ghostwriting is low and decreasing. Flanagin[26] and Wislar[19] conducted two similar cross-sectional surveys of corresponding authors of articles published in six general medicine journals in 1996[26] and in 2008.[19] The surveys, which included a core set of three journals (Annals of Internal Medicine, JAMA, the New England Journal of Medicine) at each time point and in targeted populations, used pretested questions that required an anonymous response and had response rates greater than 65%. The prevalence of ghost authoring (which included ghostwriting) was estimated to be 11.5% in 1996 and 7.9% in 2008. The prevalence of ghostwriting from these surveys was estimated to be 1.4% in 1996 and 0.2% in 2008. Hamilton and Jacobs conducted a cross-sectional survey of members of two medical writing associations in 2005, 2008, and 2011.[24, 32] The survey, which was not conducted in targeted populations, used pretested questions that required an anonymous response and had response rates from 12% to 28% of the total population surveyed (ie, not all participants

surveyed were involved in the preparation of peer-reviewed manuscripts). Findings from these surveys suggested that the percentage of association members who contributed to peer-reviewed publications and had ghostwritten at least once during the survey year had decreased from 61.8% in 2005 to 33.0% in 2011.

Six cross-sectional survey publications reported the prevalence of possible ghostwriting (ie, the prevalence of ghostwriting and ghost authoring combined).[19, 21, 22, 26, 27, 31] The prevalence of possible ghostwriting reported in these cross-sectional surveys varied from 0.9% to 24.1% of publications or authors.

Four cross-sectional survey publications reported the prevalence of ghost authoring.[20, 23, 25, 30] The prevalence of ghost authoring reported in these cross-sectional surveys varied from 0.7% to 70% of publications or authors.

The descriptive analyses of published articles on sertraline and rofecoxib did not include a prespecified definition of ghost authoring or ghostwriting;[4, 6] in these studies, possible ghostwriting or ghost authoring was assumed in publications associated with industry-sponsored support (Table 2). The descriptive analysis of industry-initiated trials in Denmark used a nonstandard definition of ghost authors; in this study ghost authors were defined as individuals, not named as authors, who were involved in writing the protocol, conducting the statistical analyses, or writing the manuscript.[29] The prevalence of possible ghostwriting in the two descriptive analyses of single drugs that did not include a prespecified definition of ghostwriting was 57% of articles on sertraline published from 1998 to 2000[4] and 69% of reviews on rofecoxib published from 1996 to 2004.[6] The prevalence of possible ghostwriting in the descriptive analysis of Danish industry-initiated trials, which used a nonstandard definition of authorship, was 75% of Danish initiated trials approved in 1994 to 1995.[29] The descriptive

analysis of noninferiority trials[28] was not considered further as the definition of ghostwriting used was consistent with disclosed medical writing assistance. In this study ghostwriters were defined as acknowledged individuals, other than authors, who contributed to the writing and were affiliated with an industry-sponsored study.[28] The prevalence of disclosed medical writing assistance in this descriptive analysis was 17.3% (101/583) of clinical trials.

Secondary publications

Publication characteristics

Of the 32 secondary publications, there were 13 review articles, 10 editorials, 5 commentaries, 3 news articles, and 1 government report, with most being published after 2008 (Table 3). Most publications cited primary sources as evidence of the prevalence of possible ghostwriting (Table 3), with the cited prevalence of possible ghostwriting varying from 6% of publications to 100% of publications involving drugs. In most secondary publications, the information on the prevalence of ghostwriting was not reported consistently compared with the cited evidence (Table 3). Misleading and mistaken information was reported in many publications that (i) did not distinguish between ghostwriting and ghost authoring, (ii) included acknowledged medical writing assistance or a combined estimate of guest authorship and ghost authorship as ghostwriting, (iii) generalized findings from publication reviews and analyses of specific datasets to wider populations of publications or industry-sponsored trials, or (iv) cited personal communications or informal surveys where the original source was unpublished and could not be verified (Table 3).

Table 3. Characteristics of Secondary Publications Citing Evidence of Possible Ghostwriting

Publication	Cited evidence	Consistent with cited source(s)	Comment
Elliot Hastings Cent Rep 2004[35]	Primary	No	Did not distinguish GW from GA
Moffat Perspect Biol Med 2007[36]	Primary	No	Did not distinguish GW from GA
Schiefe Pharmacotherapy 2009[47]	Primary	No	Did not distinguish GW from GA
MacLennan Climacteric 2010[48]	Primary	No	Did not distinguish GW from GA
Nahai Aesthet Surg J 2010[49]	Secondary	No	Did not distinguish GW from GA
Ngai Account Res 2005[50]	Primary	Yes	Did not distinguish GW from GA
Bosch EMBO Rep 2011[43]	Primary	Yes	Did not distinguish GW from GA
Wiwanitkit Am J Med 2012[51]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Krimsky Med Law 2007[52]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Langdon-Neuner Mens Sana Monogr 2008[53]	Primary & secondary	No	Did not distinguish GW from GA or GW from disclosed medical writing support
Tharyan Indian J Med Ethics 2011[45]	Primary	Yes	Did not distinguish GW from GA or GW from disclosed medical writing assistance
Paul Clin Microbiol Infect 2009[54]	Primary	Yes	Did not distinguish GW / GA from disclosed medical writing assistance
Bavdekar Lung India 2012[42]	Primary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Górski Transplant Proc 2010[55]	Primary &	No	Did not distinguish GW from GA

Publication	Cited evidence	Consistent with cited source(s)	Comment
	secondary		and generalized evidence to a wider population
Matias-Guiu Neurologia 2011[44]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
McHenry Mens Sana Monogr 2010[56]	Gov. report	No	Generalized evidence to a wider population
Healy BMJ 2004[57]	Primary & secondary	No / ND	Generalized evidence to a wider population and reported personal opinion of GW prevalence
Abbasi BMJ 2004[2]	Secondary	No	Secondary publication cited named individual
Mitrany Science Editor 2005[58]	None	ND	Cited named individual
Collier Can Med Assoc J 2009[3]	None	ND	Cited named individual
Kmietowicz BMJ 2004[59]	None	ND	Cited named individual
Matthews Wall St J 2005[1]	None	ND	Cited unpublished data
Bonita Heart Fail Clin 2011[60]	Primary	Yes	Consistent with cited source
Grassley Int J Occup Environ Health 2011[61]	Primary	Yes	Consistent with cited source
Nat Clin Pract Nephrol 2007[62]	Primary	Yes	Consistent with cited source
Editors J Urol 2008[14]	Primary	Yes	Consistent with cited source
Baethge Deutsches Arzteblatt 2009[63]	Primary	Yes	Consistent with cited source
Flanagin CSE Annual Mtg 2010[64]	Primary	Yes	Consistent with cited source
Murray Open Med 2010[65]	Primary	Yes	Consistent with cited source

Publication	Cited evidence	Consistent with cited source(s)	Comment
Moore BMJ 2004[66]	Secondary	Yes	Consistent with cited source
Hargreaves BMJ 2007[67]	Primary	Yes	Consistent with cited source
Jones Nature 2009[68]	Primary	Yes	Consistent with cited source

Abbreviations: CSE, Council of Science Editors; GA, ghost authoring; Gov, government; GW, ghostwriting; ND, not determined.

Case study of misleading and mistaken evidence

An analysis of secondary publications citing the possible prevalence of ghostwriting in sertraline publications provides an illuminating case study on how misleading or mistaken evidence can enter and remain in the medical literature. In 2003, Healy and Cattell showed that 57% (55/96) of articles on sertraline published from 1998 to 2000 were coordinated through a medical information company and only two acknowledged medical writing assistance.[4] The authors concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". In 2004 (October 14), Healy also provided evidence on the influence of the pharmaceutical industry on key groups at a United Kingdom House of Commons Health Select Committee investigation.[34] In answer to Question 197, Healy stated the following:

"My estimate is that, even in journals like the BMJ, the Lancet, the New England Journal of Medicine and JAMA, the leading journals in the field, if these articles have to do with therapeutics, with drugs, it may be worse perhaps for psychiatry than elsewhere, but I doubt it, 50% of these articles are ghostwritten. It may be higher."

Healy and Cattell's original evidence has been cited incorrectly and interchangeably with Healy's statement to the House of Commons Health Select Committee investigation (Figure 2). Although findings from the primary publication have been cited and interpreted correctly in two secondary publications,[35, 36] many secondary publications incorrectly cited Healy's original evidence and statement to the House of Commons Health Select Committee investigation (Figure 2).

DISCUSSION

This systematic review on the reported prevalence of ghostwriting in the medical literature showed that the estimates of the prevalence of ghostwriting in primary publications varied markedly and were influenced by the definitions used, the types of study designs, and the populations assessed. In addition, secondary publications often cited outdated, misleading, or mistaken evidence of the reported prevalence of ghostwriting, with many publications not distinguishing ghostwriting from ghost authoring.

Although evidence from descriptive analyses can highlight the extent of ghostwriting in single populations, evidence from well-conducted cross-sectional surveys have the potential to provide estimates of the prevalence of ghostwriting that may be generalized to the majority of peer-reviewed publications. Despite this, many of the cross-sectional surveys retrieved in this systematic review were conducted in limited populations that were not broadly representative of the peer-reviewed medical literature. In addition, many of the cross-sectional surveys did not differentiate between contributions that merited authorship from those that did not merit authorship and provided at best, an estimate of possible ghostwriting. The reported prevalence of ghostwriting, where ghostwriting was defined as undisclosed contributions that did not merit authorship, was retrieved from the two cross-sectional surveys of corresponding authors from

several general medicine journals and by cross-sectional surveys (repeated on three separate occasions from 2005 to 2011) of members of two major medical writing associations. Together the findings from these surveys suggested that the prevalence of ghostwriting has decreased in recent years. However, while the findings from these surveys may be considered more broadly representative of the peer-reviewed medical literature than surveys focused on single journals, single countries, or single subject areas, interpretation of these findings should take into account that respondents were required to retrospectively self-report potentially unethical or unprofessional behaviour.

Unethical authorship practices are a major concern and are an increasingly recognized problem in the medical literature.[37] As the findings from this systematic review suggest, some of these perceived problems may arise from the considerable, but unnecessary, confusion and disagreement surrounding the definitions of ghostwriting. As recognized by the World Association of Medical Editors, professional medical writers can have a legitimate role in assisting authors to communicate their research findings in the peer-reviewed literature.[13] Professional medical writers can and do improve the timeliness and quality of reporting and can assist investigators and industry sponsors to meet their ethical commitments to the disclosure and publishing of clinical trial results.[7, 38-40] As such, the misleading and mistaken reporting of the prevalence of ghostwriting that was evident in the secondary publications retrieved in this systematic review is disappointing. Specifically, the confusion surrounding definitions of ghost authorship and ghostwriting and the unbalanced focus on industry as the source of unethical authorship practices in the secondary publications takes attention away from the need to focus on all types of unethical contributions in peer-reviewed publications, irrespective of the source of the unethical practice.

The strengths of this systematic review are that a broad search strategy was employed with few restrictions to minimize any potential for publication or language bias. All study designs and publication types were considered and there were no restrictions on language. Although the full text or abstracts of 8 publications could not be retrieved, omission of these publications was unlikely to have biased the findings. Review of the abstracts or titles of these publications suggested that none reported numerical estimates of the prevalence of ghostwriting. The major finding of this review was the limitations of the reported evidence of ghostwriting in the medical literature. These limitations included the heterogeneity among the studies in the outcomes reported and populations investigated, the observational study designs, and the retrospective nature of data collection. Given the nature of unethical authorship practices, it may not be feasible to conduct a prospective study on ghostwriting. However, based on the findings from this review, recommendations can be made to help researchers, authors, editors, and peer-reviewers apply the same rigorous standards that are applied to the conduct and assessment of all clinical research and actively improve the quality of reporting of the evidence of unethical authorship practices.

Researchers should use a standard definition of ghostwriting so that the confusion around ghostwriters and ghost authors is not perpetuated. Ghostwriting is paid or unpaid writing assistance by individuals who generally do not merit authorship and whose contributions are not disclosed in the acknowledgments.[7] In contrast, ghost authoring is contributions to the research, data analysis, and/or writing of a manuscript by individuals who do merit authorship and whose contributions are not disclosed in the author byline.[7] For example, the descriptive analysis of Danish industry-initiated trials used a nonstandard definition of authorship, which is likely to have contributed to the very high prevalence of possible ghostwriting reported in this study (75%). In this study, Gøtzsche et al.[29] suggested that individuals who write the trial protocol, conduct the statistical analyses, or who contribute to the writing of a publication should

be included as authors. Indeed, the prevalence of ghost authorship in this study was 91% when the analyses included these individuals, irrespective of whether they had been appropriately acknowledged elsewhere. Given that the Gøtzsche et al.[29] study has been downloaded more than 3,000 times since publication[41] and cited repeatedly as evidence not only of ghost authorship but also of ghostwriting,[42-45] it would have been illuminating if the authors had included an estimate of the prevalence of ghostwriting using standard definitions for comparison. In addition to using standard definitions of ghostwriting, researchers and authors should ensure that cited evidence of ghostwriting is reported accurately without unwarranted generalizations. Publications based on personal commentary should be avoided and studies that use nonstandard definitions, specific populations, or that were conducted before a change in practice (eg, before the adoption of the Good Publication Practice guidelines for communicating company sponsored research in 2003 [GPP][46] and in 2009 [GPP2][11]) should be described in context.

Editors and peer reviewers have a duty to prevent outdated, misleading, or mistaken evidence on the prevalence of ghostwriting from being published and perpetuated. Close attention should be paid to the internal and external validity of the study, the definitions used, how the data are reported, and whether the data are interpreted within the context of current practices. When assessing a submitted manuscript on ghostwriting, editors should consider using peer reviewers with expertise in the appropriate study designs (eg, survey research) and ethical publication practices. Collectively, these actions could help prevent further questionable evidence on the prevalence of ghostwriting from being published.

In conclusion, the findings from this systematic review showed that reports of the prevalence of ghostwriting in the medical literature are limited by the varied definitions used to describe unethical authorship practices, the types of study designs employed, and the populations

assessed. To improve reporting, researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. While open and transparent debate should be encouraged, editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

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COMPETING INTERESTS

The author is a professional medical writer who is actively involved in national and international not-for-profit organizations that encourage ethical medical writing practices. The author has no relationships with any pharmaceutical company that might have an interest in the submitted work and has no other relationships that could be construed as a competing interest.

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FIGURE LEGENDS

Figure 1. Selection of publications reporting a numerical estimate of the prevalence of ghostwriting. Electronic databases were searched on 23 May 2013.

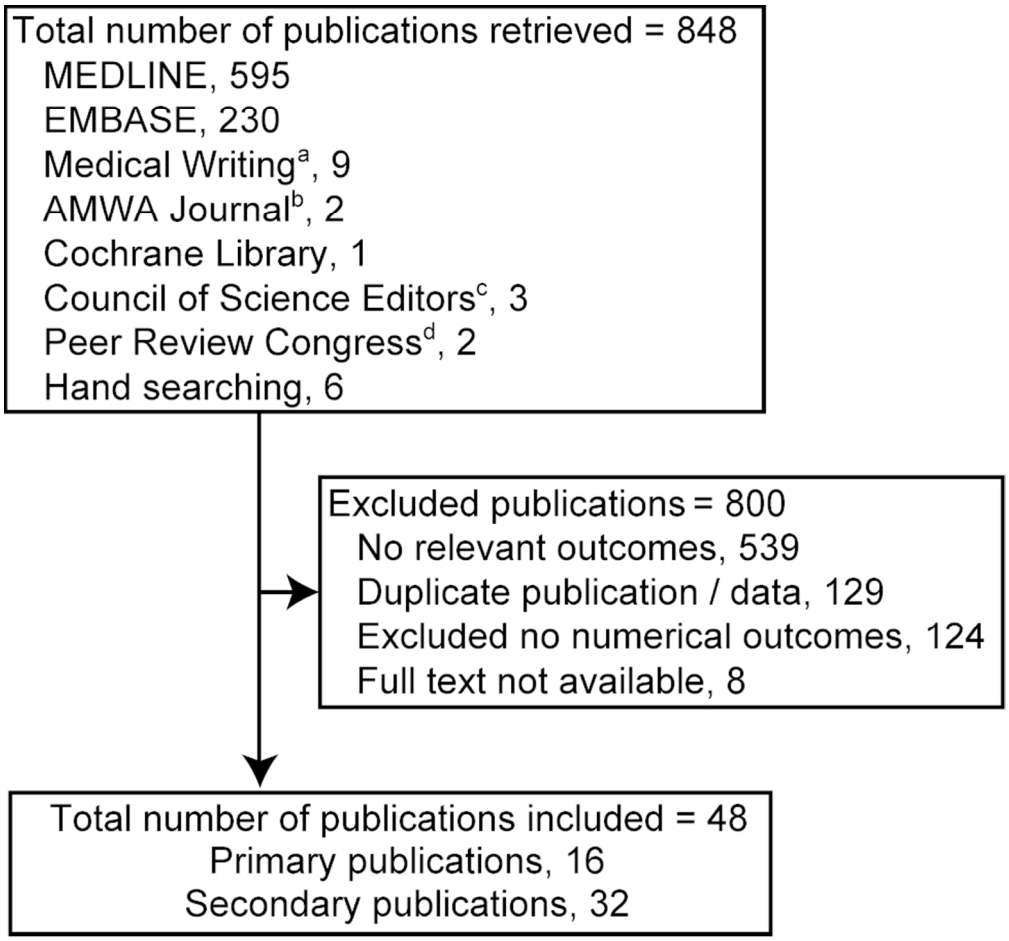
[a] Journal of the European Medical Writers Association, formerly known as The Write Stuff.

[b] Journal of the American Medical Writers Association.

[c] Council of Science Editors Annual Meetings.

[d] International Congress on Peer Review and Biomedical Publication.

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[4] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.



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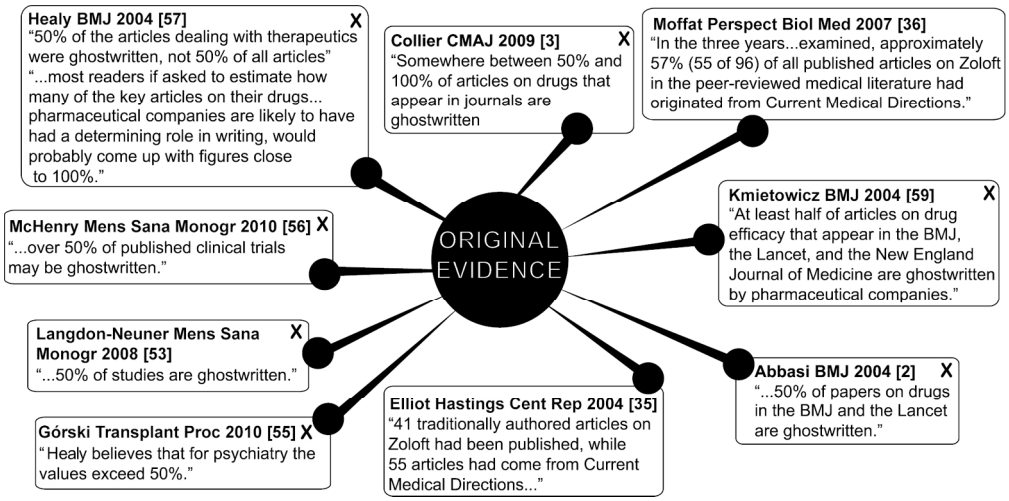
a Journal of the European Medical Writers Association, formerly known as The Write Stuff.
b Journal of the American Medical Writers Association.
c Council of Science Editors Annual Meetings.
d International Congress on Peer Review and Biomedical Publication.

82x76mm (300 x 300 DPI)



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170x83mm (300 x 300 DPI)

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2/3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	6
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	Not applicable
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6/7
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	7
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	7
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	Not applicable
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	Not applicable
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	Not applicable

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	Not applicable
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	Not applicable
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	8, Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	8-11, 15, Tables 1-3
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Not applicable
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Tables 1-3
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	Not applicable
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	Not applicable
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	Not applicable
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	20-24
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	20-22
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	22-24
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	Not applicable

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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BMJ Open

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

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TITLE

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

AUTHOR

Serina Stretton, PhD CMPP

AUTHOR'S AFFILIATION

ProScribe – Envision Pharma Group, Sydney, NSW, Australia

AUTHOR FOR CORRESPONDENCE

Serina Stretton, PhD, CMPP

ProScribe – Envision Pharma Group

18 Poulet St

Matraville, NSW, 2036

AUSTRALIA

Tel: + 612 9661 0768

Fax: + 617 5474 9156

Email: serina.stretton@EnvisionPharmaGroup.com

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Authorship, ghostwriting, systematic review

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ABSTRACT

Background: Ghostwriting of industry-sponsored articles is unethical and is perceived to be common practice.

Objective: To systematically review how evidence for the prevalence of ghostwriting is reported in the medical literature.

Data sources: MEDLINE via PubMed 1966+, EMBASE 1966+, The Cochrane Library 1988+, Medical Writing 1998+, The AMWA Journal 1986+, Council of Science Editors Annual Meetings 2007+, and the Peer Review Congress 1994+ were searched electronically (23 May 2013) using the search terms ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*.

Eligibility criteria: All publication types were considered; only publications reporting a numerical estimate of possible ghostwriting prevalence were included.

Data extraction: Two independent reviewers screened the publications; discrepancies were resolved by consensus. Data to be collected included a numerical estimate of the prevalence of possible ghostwriting (primary outcome measure), definitions of ghostwriting reported, source of the reported prevalence, publication type and year, study design, and sample population.

Results Of the 848 publications retrieved and screened for eligibility, 48 reported numerical estimates for the prevalence of possible ghostwriting. Sixteen primary publications reported findings from cross-sectional surveys or descriptive analyses of published articles; 32 secondary publications cited published or unpublished evidence. Estimates on the prevalence of possible ghostwriting in primary and secondary publications varied markedly. Primary estimates were not suitable for meta-analysis because of the various definitions of ghostwriting used, study designs, and types of populations or samples. Secondary estimates were not always reported or cited correctly or appropriately.

Conclusions Evidence for the prevalence of ghostwriting in the medical literature is limited and can be outdated, misleading, or mistaken. Researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. Editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

Strengths

- First systematic review on the reporting of the prevalence of ghostwriting in the medical literature
- A broad search strategy was employed with few restrictions to minimize any potential for publication or language bias; there were no restrictions on language.
- All study designs and publication types were considered

Limitations

- Study heterogeneity in the outcomes reported and populations investigated precluded synthesis of the data
- Retrospective and self-reported nature of the data collection increased the risk of selection bias in the studies
- Most included studies were not broadly representative of the peer-reviewed medical literature

INTRODUCTION

Ghostwriting of peer-reviewed journal publications in the medical literature is believed to be common practice.[1-3] This belief is supported, to a large extent, by highly publicized cases, primarily from the 1990s and early 2000s, of pharmaceutical companies and authors who had used ghostwriters to prepare manuscripts for publication in medical journals.[4-6] Such cases are highly unethical because the role of the commercial sponsor and any other potential conflicts of interest were hidden.

Ghostwriting occurs when writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments. This practice is distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[7] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding the two unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[8] Estimating the prevalence of ghostwriting has also been hindered by the failure of authors to distinguish ghostwriting, which is unethical, from professional medical writing support, which is ethical.[9, 10] Ghostwriters keep their involvement in a manuscript hidden, whereas professional medical writers disclose their involvement and follow ethical publication practices.[7, 9-11] Consistent with the authorship criteria recommended by the International Committee of Medical Journal Editors (ICMJE), professional medical writers who provide writing assistance and do not meet all of the ICMJE authorship criteria, should be acknowledged rather than listed as authors.[12] The prevalence of disclosed professional medical writing assistance in medical journals has been estimated to be between 6.0% and 11.0%[10, 13] and the legitimate value that this medical

writing assistance can bring to improving the quality, timeliness, and integrity of reporting in medical journals has been demonstrated.[7, 14, 15] However, the exact prevalence of ghostwriting and other forms of undisclosed contributions to papers published in medical journals is unknown.

The primary objective of this study was to conduct the first systematic review on how the prevalence of ghostwriting is reported in the medical literature. The secondary objectives were to assess the variability of the reported estimates of the prevalence of ghostwriting and investigate the source for these estimates.

MATERIALS AND METHODS

Literature search strategy

A search strategy was developed to retrieve publications from the medical literature that reported quantitative estimates of the prevalence of ghostwriting. The following databases were searched on 23 May 2013: MEDLINE via PubMed (1966+); EMBASE (1966+); The Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials (1988+); The AMWA Journal (1986+, journal of the American Medical Writers Association); Medical Writing (1998+, journal of the European Medical Writers Association, formerly known as The Write Stuff); Council of Science Editors annual meetings (2007+); and the International Congress on Peer Review and Biomedical Publication meetings (1994+). General text was searched using the following search terms: ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*. Truncation symbols and Boolean operators (AND, OR) were used wherever possible.

Two reviewers (SS, SMG) independently screened the title and abstracts of all retrieved publications using prespecified eligibility criteria; the full text of potentially eligible publications was screened to confirm eligibility for inclusion. Any discrepancies between the reviewers were resolved by consensus. The reference lists of relevant reviews and other publications were screened by hand to identify any additional publications for inclusion.

Eligibility criteria

The eligibility criteria were prespecified. Publications were included if they reported a numerical estimate of the prevalence of possible ghostwriting. Publications were excluded if they were duplicate publications from different databases or abstracts of subsequently published full-text articles, did not report any outcomes related to ghostwriting, or reported

qualitative estimate(s) of the prevalence of possible ghostwriting. No restrictions on language were included in the eligibility criteria.

Data extraction and analysis

The primary and secondary outcomes and data to be collected were prespecified. Data to be collected included publication type and year, study design and sample population, definitions of ghostwriting reported, reported prevalence of possible ghostwriting, and the source of the reported prevalence of ghostwriting. The prevalence of possible ghostwriting was reported as published. No unpublished data from the retrieved literature were reported and values for prevalence were not extrapolated from published data.

The primary outcome measure for the systematic review was a numerical estimate of the prevalence of possible ghostwriting. The reported definitions of possible ghostwriting were categorized according to the following standard definitions, irrespective of the term used to describe the practice in the publication. Undisclosed writing contributions to a manuscript were defined as (i) ghostwriting if they were described as not meriting authorship and were not listed in the acknowledgments[9, 16] and (ii) ghost authoring if they did merit authorship and were not listed in the author byline.[7] Consistent with recommendations from international medical journal editors,[12, 14] Good Publication Practice guidelines,[11] and professional medical writing associations,[17, 18] disclosed writing contributions to a manuscript that did not merit authorship and were disclosed in the acknowledgments were not categorized as ghostwriting.

The key factors considered when assessing study quality were study design, the population assessed, and how ghostwriting was defined. While there is no consensus on the best practice for reporting survey research,[19] the quality of the cross-sectional surveys was

determined by an assessment of the following factors: validation or pretesting of the survey questions, anonymity of the response, sample size, and response rate.

RESULTS

Publication selection

A total of 848 articles were retrieved from the literature search, 800 were excluded, and 48 met the eligibility criteria and were included in the systematic review (Figure 1). The main reasons for exclusion were publications not relevant to ghostwriting (n = 539), duplicate publications from different databases (n = 129), and publications not reporting a numerical estimate of the prevalence of possible ghostwriting (n = 124). The titles or abstracts of the 8 publications that were excluded because the full text or abstract could not be retrieved were reviewed; none were considered to report a numerical estimate of the prevalence of possible ghostwriting. Overall, eligible publications included 16 primary publications that reported original research on the prevalence of possible ghostwriting and 32 secondary publications that cited published or unpublished evidence of possible ghostwriting.

Primary publications

Publication characteristics

Of the 16 primary publications (Table 1), there were 13 full-text publications[4, 6, 20-30] and three conference abstracts[31-33] that reported a numerical estimate of possible ghostwriting. Twelve publications[20-28, 31-33] reported findings from cross-sectional surveys and four publications[4, 6, 29, 30] reported findings from descriptive analyses of published articles. Most cross-sectional surveys were reasonably well reported. The surveys used were not validated but most included pretested questions, required an anonymous response, were conducted in targeted populations (ie, all individuals invited to participate were involved in the preparation of peer-reviewed manuscripts), and reported a sample size and response rate. Of the cross-sectional surveys conducted in targeted populations (eg, corresponding or first authors), six had response rates greater than 50%, two had response rates less than 50%, and one did not report a response rate (Table 1). Of the cross-sectional

Table 1. Characteristics of Primary Publications Reporting Original Evidence of Ghostwriting

Publication	Population	Survey description	Data collected	N	RR
Cross-sectional surveys – authors or corresponding authors					
Flanagin 1998[27]	6 general medicine journals [a]	Self-administered, postal, anonymous, pretested, targeted	1996	809	69%
Mowatt 2002[23]	Published Cochrane reviews	Self-administered, online, anonymous, pretested, targeted	1999	362	63%
Hao 2009[32] (Abstract)	Chin Med J	Self-administered, email, not anonymous, targeted	2008 [c]	220	86%
Dotson 2011[28]	3 pharmacy journals	Self-administered, online, anonymous, pretested, targeted	2009	112	25%
Mirzazadeh 2011[24]	3 Iranian journals	Self-administered, email, not anonymous, targeted	2009-2010	NR	NR
Wislar 2011[20]	6 general medicine journals [b]	Self-administered, online, anonymous, pretested, targeted	2008	622	70%
Ghajarzadeh 2012[26]	Arch Iran Med (based on student theses)	Self-administered, email, anonymous, targeted	2005-2007	30	49%
Vinther 2012[21]	Ugeskr Laeger & Dan Med J	Self-administered, online, anonymous, pretested, targeted	2010	272	62%
Cross-sectional surveys – healthcare professionals					
Price 2000[22]	Health academic staff	Self-administered, postal, anonymous, pretested, targeted	NR	166	59%
Rees 2013[31] (Abstract)	Registered users of EPG Online	Self-administered, online, anonymous, nontargeted	NR	295	NR
Cross-sectional surveys – members of medical writing associations					
Jacobs 2009[25]	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2005 2008	843 773	28% 14%
Hamilton	EMWA / AMWA	Self-administered, online,	2011	620	12% [d]

2012[33] (Abstract)	members	anonymous, pretested, nontargeted			
Publication reviews and descriptive analysis					
Healy 2003[4]	Articles on sertraline	NA	1998-2000	96	NA
Gøtzsche 2007[30]	Articles on Danish industry-initiated trials approved 1994-1995	NA	NR	44	NA
Ross 2008[6]	Reviews on rofecoxib associated with Merck support	NA	1996-2004	72	NA
Suda 2011[29]	Noninferiority clinical trials	NA	1989-2009	583	NA

[a] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, American Journal of Cardiology, American Journal of Medicine, American Journal of Obstetrics and Gynecology.

[b] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, Lancet, Nature Medicine, PLoS Medicine.

[c] Submission date.

[d] Personal communication, C. Hamilton.

Abbreviations: AMWA, American Medical Writers Association; EMWA, European Medical Writers Association; NA, not applicable; NR, not reported; RR, response rate.

surveys conducted in nontargeted populations (eg, individuals invited to participate who may or may not have been involved in providing medical writing assistance for peer-reviewed publications), three had low (12% to 28%) response rates and one did not report a response rate (Table 1).

Not all cross-sectional surveys were broadly representative of the peer-reviewed medical literature. The cross-sectional surveys were conducted in single populations of academic staff or healthcare professionals[22, 31], members of medical writing associations[25, 33], or

corresponding or first authors. The corresponding or first authors were surveyed from single journals (Cochrane reviews, Chinese Medical Journal, Archives of Iranian Medicine),[23, 26, 32] two Danish journals,[21] three Iranian journals,[24] and three pharmacy journals.[28] Two cross-sectional surveys of corresponding authors were conducted in six general medicine journals.[20, 27]

The descriptive analyses of published articles were conducted in single populations and included an analysis of sertraline publications from 1998 to 2000,[4] rofecoxib reviews from 1996 to 2004,[6] publications from industry-initiated trials in Denmark from 1994 to 1995,[30] and from published noninferiority trials from 1989 to 2009.[29]

Reported prevalence of ghostwriting

The reported prevalence of possible ghostwriting in the primary publications varied markedly and were not suitable for meta-analysis because of the different populations assessed, the different methods used to generate the estimates, and the various definitions that were used (Tables 1 and 2). All cross-sectional surveys reported a definition of ghostwriting or ghost authoring, but most definitions did not differentiate contributions that merited authorship from those that did not merit authorship (Table 2).

Four cross-sectional surveys reported the prevalence of ghostwriting where the definition provided could be categorized as undisclosed contributions that do not merit authorship.[20, 25, 27, 33] Findings from these surveys, which were repeated at different time points, suggested that the prevalence of ghostwriting is low and decreasing. Flanagin[27] and Wislar[20] conducted two similar cross-sectional surveys of corresponding authors of articles published in six general medicine journals in 1996[27] and in 2008.[20] The surveys, which included a core set of three journals (Annals of Internal Medicine, JAMA, the New England Journal of Medicine) at each time point and in targeted populations, used pretested

Table 2. Primary Publications Reporting Estimates of the Prevalence of Possible Ghostwriting

Publication	Measure of Possible Ghostwriting Reported by Authors	Estimate % (n)
Cross-sectional surveys reporting ghostwriting		
Flanagin 1998[27]	Unnamed individual who participated in the writing	1.4% (11/809) of articles
Wislar 2011[20]	Unnamed individual who participated in the writing	0.2% [a] (1/622) of articles
Jacobs 2009, Hamilton 2012[25, 33]	Undisclosed medical writing assistance not qualifying for authorship	2005: 61.8% (NR) 2009: 41.7% (NR) 2011: 33.0% (NR) of articles [b]
Cross-sectional surveys reporting combined ghost authoring and ghostwriting		
Flanagin 1998[27]	Failure to name, as an author, individuals who made substantial contributions to the research or writing or an unidentified medical writer	11.5% (93/809) of articles
Price 2000[22]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	24.1% (40/166) of authors
Mowatt 2002[23]	Individual merited authorship or had assisted with drafting but not listed as an author or acknowledged	8.8% (32/362) of articles
Hao 2009[32]	English-language speakers assisted with writing but not identified as authors or acknowledged	10.4% (NR) of authors
Dotson 2011[28]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	0.9% (1/112) of articles
Wislar 2011[20]	Failure to name, as an author, individuals who made substantial contributions to the research or writing of the article or an unnamed individual who participated in the writing	7.9% (49/622) of articles
Cross-sectional surveys reporting ghost authoring		
Mirzazadeh 2011[24]	Failure to name, as an author, individuals who made substantial contributions to the research	21.4% (25/NR) of authors
Ghajarzadeh	Failure to name, as an author, students who made substantial	0.7% (2/296) of

2012[26]	contributions to the research [c]	articles
Vinther 2012[21]	Individual merited authorship but not listed as an author	2.4% (6/245) of articles
Rees 2013[31]	Individual merited authorship but not listed as an author	70% (NR/202) of published authors
Publication reviews and descriptive analysis reporting possible ghost authoring or ghostwriting		
Healy 2003[4]	Published articles coordinated by a medical information company, including acknowledged medical writing support [d]	57.3% (55/96) of articles
Gøtzsche 2007[30]	Individuals who wrote the trial protocol, conducted the statistical analyses, or wrote the manuscript but were not listed as authors, not members of a study group or steering committee, or not disclosed in an acknowledgment	75.0% (33/44) of trials
Ross 2008[6]	Published reviews associated with Merck support and with a single external author [e]	69.4% (50/72) of reviews

[a] Available as online supplementary data.

[b] Values represent the mean weighted percentage of publications that were ghostwritten by respondents. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

[c] Students were classified as ghostwriters if the student was not named as an author and if the results reported in the publications were based on the results of their theses.

[d] Authors conclude data provide quantification of the possible extent of ghostwriting based on a single drug. Of the 55 published articles that were coordinated through a medical information company, 2 included medical writing assistance that was acknowledged in the published article.

[e] Published review articles had been identified from correspondence between Merck and a medical publishing company, from Merck publication status reports, or were affiliated with an author named within the correspondence or publication status reports. The authors did not report whether medical writing assistance was acknowledged in the published article.

Abbreviations: NR, not reported.

questions that required an anonymous response and had response rates greater than 65%. The prevalence of ghost authoring (which included ghostwriting) was estimated to be 11.5% in 1996 and 7.9% in 2008. The prevalence of ghostwriting from these surveys was estimated to be 1.4% in 1996 and 0.2% in 2008. Hamilton and Jacobs conducted a cross-sectional survey of members of two medical writing associations in 2005, 2008, and 2011.[25, 33] The survey, which was not conducted in targeted populations, used pretested questions that required an anonymous response and had response rates from 12% to 28% of the total population surveyed (ie, not all participants surveyed were involved in the preparation of peer-reviewed manuscripts). Findings from these surveys showed that the mean weighted percentage of publications that were ghostwritten by respondents had decreased from 61.8% in 2005 to 33.0% in 2011. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

Six cross-sectional survey publications reported the prevalence of possible ghostwriting (ie, the prevalence of ghostwriting and ghost authoring combined).[20, 22, 23, 27, 28, 32] The prevalence of possible ghostwriting reported in these cross-sectional surveys varied from 0.9% to 24.1% of publications or authors.

Four cross-sectional survey publications reported the prevalence of ghost authoring.[21, 24, 26, 31] The prevalence of ghost authoring reported in these cross-sectional surveys varied from 0.7% to 70% of publications or authors.

The descriptive analyses of published articles on sertraline and rofecoxib did not include a prespecified definition of ghost authoring or ghostwriting;[4, 6] in these studies, possible ghostwriting or ghost authoring was assumed in publications associated with industry-sponsored support (Table 2). The descriptive analysis of industry-initiated trials in Denmark used a nonstandard definition of ghost authors; in this study ghost authors were defined as individuals, not named as authors, who were involved in writing the protocol, conducting the

statistical analyses, or writing the manuscript.[30] The prevalence of possible ghostwriting in the two descriptive analyses of single drugs that did not include a prespecified definition of ghostwriting was 57% of articles on sertraline published from 1998 to 2000[4] and 69% of reviews on rofecoxib published from 1996 to 2004.[6] The prevalence of possible ghostwriting in the descriptive analysis of Danish industry-initiated trials, which used a nonstandard definition of authorship, was 75% of Danish initiated trials approved in 1994 to 1995.[30] The descriptive analysis of noninferiority trials[29] was not considered further as the definition of ghostwriting used was consistent with disclosed medical writing assistance. In this study ghostwriters were defined as acknowledged individuals, other than authors, who contributed to the writing and were affiliated with an industry-sponsored study.[29] The prevalence of disclosed medical writing assistance in this descriptive analysis was 17.3% (101/583) of clinical trials.

Secondary publications

Publication characteristics

Of the 32 secondary publications, there were 13 review articles, 10 editorials, 5 commentaries, 3 news articles, and 1 government report, with most being published after 2008 (Table 3). Most publications cited primary sources as evidence of the prevalence of possible ghostwriting (Table 3), with the cited prevalence of possible ghostwriting varying from 6% of publications to 100% of publications involving drugs. In most secondary publications, the information on the prevalence of ghostwriting was not reported consistently compared with the cited evidence (Table 3). Misleading and mistaken information was reported in many publications that (i) did not distinguish between ghostwriting and ghost authoring, (ii) included acknowledged medical writing assistance or a combined estimate of guest authorship and ghost authorship as ghostwriting, (iii) generalized findings from publication reviews and analyses of specific datasets to wider populations of publications or

Table 3. Characteristics of Secondary Publications Citing Evidence of Possible Ghostwriting

Publication	Cited evidence	Consistent with cited source(s)	Comment
Elliot Hastings Cent Rep 2004[35]	Primary	No	Did not distinguish GW from GA
Moffat Perspect Biol Med 2007[36]	Primary	No	Did not distinguish GW from GA
Schiefe Pharmacotherapy 2009[47]	Primary	No	Did not distinguish GW from GA
MacLennan Climacteric 2010[48]	Primary	No	Did not distinguish GW from GA
Nahai Aesthet Surg J 2010[49]	Secondary	No	Did not distinguish GW from GA
Ngai Account Res 2005[50]	Primary	Yes	Did not distinguish GW from GA
Bosch EMBO Rep 2011[43]	Primary	Yes	Did not distinguish GW from GA
Wiwanitkit Am J Med 2012[51]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Krimsky Med Law 2007[52]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Langdon-Neuner Mens Sana Monogr 2008[53]	Primary & secondary	No	Did not distinguish GW from GA or GW from disclosed medical writing support
Tharyan Indian J Med Ethics 2011[45]	Primary	Yes	Did not distinguish GW from GA or GW from disclosed medical writing assistance
Paul Clin Microbiol Infect 2009[54]	Primary	Yes	Did not distinguish GW / GA from disclosed medical writing assistance
Bavdekar Lung India 2012[42]	Primary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Górski Transplant Proc 2010[55]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population

Matias-Guiu Neurologia 2011[44]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
McHenry Mens Sana Monogr 2010[56]	Gov. report	No	Generalized evidence to a wider population
Healy BMJ 2004[57]	Primary & secondary	No / ND	Generalized evidence to a wider population and reported personal opinion of GW prevalence
Abbasi BMJ 2004[2]	Secondary	No	Secondary publication cited named individual
Mitrany Science Editor 2005[58]	None	ND	Cited named individual
Collier Can Med Assoc J 2009[3]	None	ND	Cited named individual
Kmietowicz BMJ 2004[59]	None	ND	Cited named individual
Matthews Wall St J 2005[1]	None	ND	Cited unpublished data
Bonita Heart Fail Clin 2011[60]	Primary	Yes	Consistent with cited source
Grassley Int J Occup Environ Health 2011[61]	Primary	Yes	Consistent with cited source
Nat Clin Pract Nephrol 2007[62]	Primary	Yes	Consistent with cited source
Editors J Urol 2008[15]	Primary	Yes	Consistent with cited source
Baethge Deutsches Arzteblatt 2009[63]	Primary	Yes	Consistent with cited source
Flanagin CSE Annual Mtg 2010[64]	Primary	Yes	Consistent with cited source
Murray Open Med 2010[65]	Primary	Yes	Consistent with cited source
Moore BMJ 2004[66]	Secondary	Yes	Consistent with cited source
Hargreaves BMJ 2007[67]	Primary	Yes	Consistent with cited source
Jones Nature 2009[68]	Primary	Yes	Consistent with cited source

Abbreviations: CSE, Council of Science Editors; GA, ghost authoring; Gov, government; GW, ghostwriting; ND, not determined.

industry-sponsored trials, or (iv) cited personal communications or informal surveys where the original source was unpublished and could not be verified (Table 3).

Case study of misleading and mistaken evidence

An analysis of secondary publications citing the possible prevalence of ghostwriting in sertraline publications provides an illuminating case study on how misleading or mistaken evidence can enter and remain in the medical literature. In 2003, Healy and Cattell showed that 57% (55/96) of articles on sertraline published from 1998 to 2000 were coordinated through a medical information company and only two acknowledged medical writing assistance.[4] The authors concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". In 2004 (October 14), Healy also provided evidence on the influence of the pharmaceutical industry on key groups at a United Kingdom House of Commons Health Select Committee investigation.[34] In answer to Question 197, Healy stated the following:

"My estimate is that, even in journals like the BMJ, the Lancet, the New England Journal of Medicine and JAMA, the leading journals in the field, if these articles have to do with therapeutics, with drugs, it may be worse perhaps for psychiatry than elsewhere, but I doubt it, 50% of these articles are ghostwritten. It may be higher."

Healy and Cattell's original evidence has been cited incorrectly and interchangeably with Healy's statement to the House of Commons Health Select Committee investigation (Figure 2). Although findings from the primary publication have been cited and interpreted correctly in two secondary publications,[35, 36] many secondary publications incorrectly cited Healy's original evidence and statement to the House of Commons Health Select Committee investigation (Figure 2).

DISCUSSION

This systematic review on the reported prevalence of ghostwriting in the medical literature showed that the estimates of the prevalence of ghostwriting in primary publications varied markedly and were influenced by the definitions used, the types of study designs, and the type of population or sample assessed. In addition, secondary publications often cited outdated, misleading, or mistaken evidence of the reported prevalence of ghostwriting, with many publications not distinguishing ghostwriting from ghost authoring.

Although evidence from descriptive analyses can highlight the extent of ghostwriting in single populations, evidence from well-conducted cross-sectional surveys have the potential to provide estimates of the prevalence of ghostwriting that may be generalized to the majority of peer-reviewed publications. Despite this, many of the cross-sectional surveys retrieved in this systematic review were conducted in limited populations that were not broadly representative of the peer-reviewed medical literature. In addition, many of the cross-sectional surveys did not differentiate between contributions that merited authorship from those that did not merit authorship and provided at best, an estimate of possible ghostwriting. The reported prevalence of ghostwriting, where ghostwriting was defined as undisclosed contributions that did not merit authorship, was retrieved from the two cross-sectional surveys of corresponding authors from several general medicine journals and by cross-sectional surveys (repeated on three separate occasions from 2005 to 2011) of members of two major medical writing associations. Together the findings from these surveys suggested that the prevalence of ghostwriting has decreased in recent years. However, while the findings from these surveys may be considered more broadly representative of the peer-reviewed medical literature than surveys focused on single journals, single countries, or single subject areas, interpretation of these findings should take

into account that respondents were required to retrospectively self-report potentially unethical or unprofessional behaviour.

Unethical authorship practices are a major concern and are an increasingly recognized problem in the medical literature.[37] As the findings from this systematic review suggest, some of these perceived problems may arise from the considerable, but unnecessary, confusion and disagreement surrounding the definitions of ghostwriting. As recognized by the World Association of Medical Editors, professional medical writers can have a legitimate role in assisting authors to communicate their research findings in the peer-reviewed literature.[14] Professional medical writers can and do improve the timeliness and quality of reporting and can assist investigators and industry sponsors to meet their ethical commitments to the disclosure and publishing of clinical trial results.[7, 38-40] As such, the misleading and mistaken reporting of the prevalence of ghostwriting that was evident in the secondary publications retrieved in this systematic review is disappointing. Specifically, the confusion surrounding definitions of ghost authorship and ghostwriting and the unbalanced focus on industry as the source of unethical authorship practices in the secondary publications takes attention away from the need to focus on all types of unethical contributions in peer-reviewed publications, irrespective of the source of the unethical practice.

The strengths of this systematic review are that a broad search strategy was employed with few restrictions to minimize any potential for publication or language bias. All study designs and publication types were considered and there were no restrictions on language. Although the full text or abstracts of 8 publications could not be retrieved, omission of these publications was unlikely to have biased the findings. Review of the abstracts or titles of these publications suggested that none reported numerical estimates of the prevalence of ghostwriting. The major finding of this review was the limitations of the reported evidence of ghostwriting in the medical literature. These limitations included the heterogeneity among the

studies in the outcomes reported and populations investigated, the observational study designs, and the retrospective nature of data collection. Given the nature of unethical authorship practices, it may not be feasible to conduct a prospective study on ghostwriting. However, based on the findings from this review, recommendations can be made to help researchers, authors, editors, and peer-reviewers apply the same rigorous standards that are applied to the conduct and assessment of all clinical research and actively improve the quality of reporting of the evidence of unethical authorship practices.

Researchers should use a standard definition of ghostwriting so that the confusion around ghostwriters and ghost authors is not perpetuated. Ghostwriting is paid or unpaid writing assistance by individuals who generally do not merit authorship and whose contributions are not disclosed in the acknowledgments.[7] In contrast, ghost authoring is contributions to the research, data analysis, and/or writing of a manuscript by individuals who do merit authorship and whose contributions are not disclosed in the author byline.[7] For example, the descriptive analysis of Danish industry-initiated trials used a nonstandard definition of authorship, which is likely to have contributed to the very high prevalence of possible ghostwriting reported in this study (75%). In this study, Gøtzsche et al.[30] suggested that individuals who write the trial protocol, conduct the statistical analyses, or who contribute to the writing of a publication should be included as authors. Indeed, the prevalence of ghost authorship in this study was 91% when the analyses included these individuals, irrespective of whether they had been appropriately acknowledged elsewhere. Given that the Gøtzsche et al.[30] study has been downloaded more than 3,000 times since publication[41] and cited repeatedly as evidence not only of ghost authorship but also of ghostwriting,[42-45] it would have been illuminating if the authors had included an estimate of the prevalence of ghostwriting using standard definitions for comparison. In addition to using standard definitions of ghostwriting, researchers and authors should ensure that cited evidence of ghostwriting is reported accurately without unwarranted generalizations. Publications based on personal commentary should be avoided and studies that use nonstandard definitions,

specific populations, or that were conducted before a change in practice (eg, before the adoption of the Good Publication Practice guidelines for communicating company sponsored research in 2003 [GPP][46] and in 2009 [GPP2][11]) should be described in context.

Editors and peer reviewers have a duty to prevent outdated, misleading, or mistaken evidence on the prevalence of ghostwriting from being published and perpetuated. Close attention should be paid to the internal and external validity of the study, the definitions used, how the data are reported, and whether the data are interpreted within the context of current practices. When assessing a submitted manuscript on ghostwriting, editors should consider using peer reviewers with expertise in the appropriate study designs (eg, survey research) and ethical publication practices. Collectively, these actions could help prevent further questionable evidence on the prevalence of ghostwriting from being published.

In conclusion, the findings from this systematic review showed that reports of the prevalence of ghostwriting in the medical literature are limited by the varied definitions used to describe unethical authorship practices, the types of study designs employed, and the populations assessed. To improve reporting, researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. While open and transparent debate should be encouraged, editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

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FIGURE LEGENDS

Figure 1. Selection of publications reporting a numerical estimate of the prevalence of ghostwriting. Electronic databases were searched on 23 May 2013.

[a] Journal of the European Medical Writers Association, formerly known as The Write Stuff.

[b] Journal of the American Medical Writers Association.

[c] Council of Science Editors Annual Meetings.

[d] International Congress on Peer Review and Biomedical Publication.

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[4] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.

TITLE

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

AUTHOR

Serina Stretton, PhD CMPP

AUTHOR'S AFFILIATION

ProScribe – Envision Pharma Group ~~Medical Communications~~, ~~Noosaville, Queensland~~Sydney, NSW, Australia

AUTHOR FOR CORRESPONDENCE

Serina Stretton, PhD, CMPP

ProScribe – Envision Pharma Group~~Medical Communications~~

18 Poulet St

Matraville, NSW, 2036

AUSTRALIA

Tel: + 612 9661 0768

Fax: + 61~~2~~ 7 5474 9156~~9661 0766~~

Email: serina.stretton@EnvisionPharmaGroup.com~~ss@proscribe.com.au~~

KEYWORDS

Authorship, ghostwriting, systematic review

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ABSTRACT

Background: Ghostwriting of industry-sponsored articles is unethical and is perceived to be common practice ~~in the medical literature~~.

Objective: To ~~he conduct a~~ systematically review ~~on~~ how evidence for the prevalence of ghostwriting is reported in the medical literature.

Data sources: MEDLINE via PubMed 1966+, EMBASE 1966+, The Cochrane Library 1988+, Medical Writing 1998+, The AMWA Journal 1986+, Council of Science Editors Annual Meetings 2007+, and the Peer Review Congress 1994+ were searched electronically (23 May 2013) using the search terms ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*.

Eligibility criteria: All publication types were considered; only publications reporting a numerical estimate of possible ghostwriting prevalence were included.

Data extraction: Two independent reviewers screened the publications; discrepancies were resolved by consensus. Data to be collected included a numerical estimate of the prevalence of possible ghostwriting (primary outcome measure), definitions of ghostwriting reported, source of the reported prevalence, publication type and year, study design, and sample population.

Results Of the 848 publications retrieved and screened for eligibility, 48 reported numerical estimates for the prevalence of possible ghostwriting ~~and were analyzed further~~. Sixteen primary publications reported findings from cross-sectional surveys or descriptive analyses of published articles; ~~and~~ 32 secondary publications cited published or unpublished evidence. Estimates on the prevalence of possible ghostwriting in primary and secondary publications varied markedly. Primary estimates were not suitable for meta-analysis because of the various ~~. Estimates were influenced by the~~ definitions of ghostwriting used, ~~the~~ study designs, and types of populations or samples ~~assessed~~. Secondary estimates were not

always reported or , and whether evidence from primary publications was cited correctly or appropriately.

Conclusions **Evidence for the prevalence of ghostwriting** Estimates of ghostwriting report ed in the medical literature is limited and can be outdated, misleading, or mistaken. Researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. Editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

Strengths

- First systematic review on the reporting of the prevalence of ghostwriting in the medical literature
- A broad search strategy was employed with few restrictions to minimize any potential for publication or language bias; there were no restrictions on language.
- All study designs and publication types were considered

Limitations

- Study heterogeneity in the outcomes reported and populations investigated precluded synthesis of the data
- Retrospective and self-reported nature of the data collection increased the risk of selection bias in the studies
- Most included studies were not broadly representative of the peer-reviewed medical literature

INTRODUCTION

Ghostwriting of peer-reviewed journal publications in the medical literature is believed to be common practice.[1-3] This belief is supported, to a large extent, by highly publicized cases, primarily from the 1990s and early 2000s, of pharmaceutical companies and authors who had used ghostwriters to prepare manuscripts for publication in medical journals.[4-6] Such cases are highly unethical because the role of the commercial sponsor and any other potential conflicts of interest were hidden.

Ghostwriting occurs when writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments. This practice is distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[7] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding the two unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[8] Estimating the prevalence of ghostwriting has also been hindered by the failure of authors to distinguish ghostwriting, which is unethical, from professional medical writing support, which is ethical.[9, 10] Ghostwriters keep their involvement in a manuscript hidden, whereas professional medical writers disclose their involvement and follow ethical publication practices.[7, 9-11] Consistent with the authorship criteria recommended by the International Committee of Medical Journal Editors (ICMJE), professional medical writers who provide writing assistance and do not meet all of the ICMJE authorship criteria, should be acknowledged rather than listed as authors.[12] The prevalence of disclosed professional medical writing assistance in medical journals has been estimated to be between 6.0% and 11.0%[10, 13] and the legitimate value that this medical

writing assistance can bring to improving the quality, timeliness, and integrity of reporting in medical journals has been demonstrated.[7, 14, 15] However, the exact prevalence of ghostwriting and other forms of undisclosed contributions to papers published in medical journals is unknown.

The primary objective of this study was to conduct the first systematic review on how the prevalence of ghostwriting is reported in the medical literature. The secondary objectives were to assess the variability of the reported estimates of the prevalence of ghostwriting and investigate the source for these estimates.

MATERIALS AND METHODS

Literature search strategy

A search strategy was developed to retrieve publications from the medical literature that reported quantitative estimates of the prevalence of ghostwriting. The following databases were searched on 23 May 2013: MEDLINE via PubMed (1966+); EMBASE (1966+); The Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials (1988+); The AMWA Journal (1986+, journal of the American Medical Writers Association); Medical Writing (1998+, journal of the European Medical Writers Association, formerly known as The Write Stuff); Council of Science Editors annual meetings (2007+); and the International Congress on Peer Review and Biomedical Publication meetings (1994+). General text was searched using the following search terms: ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*. Truncation symbols and Boolean operators (AND, OR) were used wherever possible.

Two reviewers (SS, SMG) independently screened the title and abstracts of all retrieved publications using prespecified eligibility criteria; the full text of potentially eligible publications was screened to confirm eligibility for inclusion. Any discrepancies between the reviewers were resolved by consensus. The reference lists of relevant reviews and other publications were screened by hand to identify any additional publications for inclusion.

Eligibility criteria

The eligibility criteria were prespecified. Publications were included if they reported a numerical estimate of the prevalence of possible ghostwriting. Publications were excluded if they were duplicate publications from different databases or abstracts of subsequently published full-text articles, did not report any outcomes related to ghostwriting, or reported

qualitative estimate(s) of the prevalence of possible ghostwriting. No restrictions on language were included in the eligibility criteria.

Data extraction and analysis

The primary and secondary outcomes and d~~D~~ata to be collected were prespecified. Data to be collected ~~and~~ included publication type and year, study design and sample population, definitions of ghostwriting reported, reported prevalence of possible ghostwriting, and the source of the reported prevalence of ghostwriting. The prevalence of possible ghostwriting was reported as published. No unpublished data from the retrieved literature were reported and values for prevalence were not extrapolated from published data.

The primary outcome measure for the systematic review was a numerical estimate of the prevalence of possible ghostwriting. The reported definitions of possible ghostwriting were categorized according to the following standard definitions, irrespective of the term used to describe the practice in the publication. Undisclosed writing contributions to a manuscript were defined as (i) ghostwriting if they were described as not meriting authorship and were not listed in the acknowledgments[9, 16] and (ii) ghost authoring if they did merit authorship and were not listed in the author byline.[7] Consistent with recommendations from international medical journal editors,[12, 14] Good Publication Practice guidelines,[11] and professional medical writing associations,[17, 18] disclosed writing contributions to a manuscript that did not merit authorship and were disclosed in the acknowledgments were not categorized as ghostwriting.

The key factors considered when assessing study quality were study design, the population assessed, and how ghostwriting was defined. While there is no consensus on the best practice for reporting survey research,[19] the quality of the cross-sectional surveys was

determined by an assessment of the following factors: validation or pretesting of the survey questions, anonymity of the response, sample size, and response rate.

RESULTS

Publication selection

A total of 848 articles were retrieved from the literature search, 800 were excluded, and 48 met the eligibility criteria and were included in the systematic review (Figure 1). The main reasons for exclusion were publications not relevant to ghostwriting (n = 539), duplicate publications from different databases (n = 129), and publications not reporting a numerical estimate of the prevalence of possible ghostwriting (n = 124). The titles or abstracts of the 8 publications that were excluded because the full text or abstract could not be retrieved were reviewed; none were considered to report a numerical estimate of the prevalence of possible ghostwriting. Overall, eligible publications included 16 primary publications that reported original research on the prevalence of possible ghostwriting and 32 secondary publications that cited published or unpublished evidence of possible ghostwriting.

Primary publications

Publication characteristics

Of the 16 primary publications (Table 1), there were 13 full-text publications[4, 6, 20-30] and three conference abstracts[31-33] that reported a numerical estimate of possible ghostwriting. Twelve publications[20-28, 31-33] reported findings from cross-sectional surveys and four publications[4, 6, 29, 30] reported findings from descriptive analyses of published articles. ~~While there is no consensus on the best practice for reporting survey research,[33]~~M ~~m~~ost cross-sectional surveys were reasonably well reported. The surveys used were not validated but most included pretested questions, required an anonymous response, were conducted in targeted populations (ie, all individuals invited to participate were involved in the preparation of peer-reviewed manuscripts), and reported a sample size and response rate. Of the cross-sectional surveys conducted in targeted populations (eg,

corresponding or first authors), six had response rates greater than 50%, two had response rates less than 50%, and one did not report a response rate (Table 1). Of the cross-sectional

Table 1. Characteristics of Primary Publications Reporting Original Evidence of Ghostwriting

Publication	Population	Survey description	Data collected	N	RR
Cross-sectional surveys – authors or corresponding authors					
Flanagin 1998[27]	6 general medicine journals [a]	Self-administered, postal, anonymous, pretested, targeted	1996	809	69%
Mowatt 2002[23]	Published Cochrane reviews	Self-administered, online, anonymous, pretested, targeted	1999	362	63%
Hao 2009[32] (Abstract)	Chin Med J	Self-administered, email, not anonymous, targeted	2008 [c]	220	86%
Dotson 2011[28]	3 pharmacy journals	Self-administered, online, anonymous, pretested, targeted	2009	112	25%
Mirzazadeh 2011[24]	3 Iranian journals	Self-administered, email, not anonymous, targeted	2009-2010	NR	NR
Wislar 2011[20]	6 general medicine journals [b]	Self-administered, online, anonymous, pretested, targeted	2008	622	70%
Ghajarzadeh 2012[26]	Arch Iran Med (based on student theses)	Self-administered, email, anonymous, targeted	2005-2007	30	49%
Vinther 2012[21]	Ugeskr Laeger & Dan Med J	Self-administered, online, anonymous, pretested, targeted	2010	272	62%
Cross-sectional surveys – healthcare professionals					
Price 2000[22]	Health academic staff	Self-administered, postal, anonymous, pretested, targeted	NR	166	59%
Rees 2013[31] (Abstract)	Registered users of EPG Online	Self-administered, online, anonymous, nontargeted	NR	295	NR
Cross-sectional surveys – members of medical writing associations					
Jacobs 2009[25]	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2005 2008	843 773	28% 14%
Hamilton	EMWA / AMWA	Self-administered, online,	2011	620	12% [d]

2012[33] (Abstract)	members	anonymous, pretested, nontargeted			
Publication reviews and descriptive analysis					
Healy 2003[4]	Articles on sertraline	NA	1998-2000	96	NA
Gøtzsche 2007[30]	Articles on Danish industry-initiated trials approved 1994-1995	NA	NR	44	NA
Ross 2008[6]	Reviews on rofecoxib associated with Merck support	NA	1996-2004	72	NA
Suda 2011[29]	Noninferiority clinical trials	NA	1989-2009	583	NA

[a] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, American Journal of Cardiology, American Journal of Medicine, American Journal of Obstetrics and Gynecology.

[b] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, Lancet, Nature Medicine, PLoS Medicine.

[c] Submission date.

[d] Personal communication, C. Hamilton.

Abbreviations: AMWA, American Medical Writers Association; EMWA, European Medical Writers Association; NA, not applicable; NR, not reported; RR, response rate.

surveys conducted in nontargeted populations (eg, individuals invited to participate who may or may not have been involved in providing medical writing assistance for peer-reviewed publications), three had low (12% to 28%) response rates and one did not report a response rate (Table 1).

Not all cross-sectional surveys were broadly representative of the peer-reviewed medical literature. The cross-sectional surveys were conducted in single populations of academic staff or healthcare professionals[22, 31], members of medical writing associations[25, 33], or

corresponding or first authors. The corresponding or first authors were surveyed from single journals (Cochrane reviews, Chinese Medical Journal, Archives of Iranian Medicine),[23, 26, 32] two Danish journals,[21] three Iranian journals,[24] and three pharmacy journals.[28] Two cross-sectional surveys of corresponding authors were conducted in six general medicine journals.[20, 27]

The descriptive analyses of published articles were conducted in single populations and included an analysis of sertraline publications from 1998 to 2000,[4] rofecoxib reviews from 1996 to 2004,[6] publications from industry-initiated trials in Denmark from 1994 to 1995,[30] and from published noninferiority trials from 1989 to 2009.[29]

Reported prevalence of ghostwriting

The reported prevalence of possible ghostwriting in the primary publications varied markedly and were not suitable for meta-analysis~~and was difficult to compare~~ because of the different populations assessed, the different methods used to generate the estimates, and the various definitions that were used (Tables 1 and 2). All cross-sectional surveys reported a definition of ghostwriting or ghost authoring, but most definitions did not differentiate contributions that merited authorship from those that did not merit authorship (Table 2).

Four cross-sectional surveys reported the prevalence of ghostwriting where the definition provided could be categorized as undisclosed contributions that do not merit authorship.[20, 25, 27, 33] Findings from these surveys, which were repeated at different time points, suggested that the prevalence of ghostwriting is low and decreasing. Flanagin[27] and Wislar[20] conducted two similar cross-sectional surveys of corresponding authors of articles published in six general medicine journals in 1996[27] and in 2008.[20] The surveys, which included a core set of three journals (Annals of Internal Medicine, JAMA, the New England Journal of Medicine) at each time point and in targeted populations, used pretested

Table 2. Primary Publications Reporting Estimates of the Prevalence of Possible Ghostwriting

Publication	Measure of Possible Ghostwriting Reported ~~measure~~by Authors	Estimate % (n)
Cross-sectional surveys reporting ghostwriting		
Flanagin 1998[27]	Unnamed individual who participated in the writing	1.4% (11/809) of articles
Wislar 2011[20]	Unnamed individual who participated in the writing	0.2%[a] (1/622) of articles
Jacobs 2009, Hamilton 2012[25, 33]	Undisclosed medical writing assistance not qualifying for authorship	2005: 61.8% (NR) 2009: 41.7% (NR) 2011: 33.0% (NR) of ~~writers~~articles[b]
Cross-sectional surveys reporting combined ghost authoring and ghostwriting		
Flanagin 1998[27]	Failure to name, as an author, individuals who made substantial contributions to the research or writing or an unidentified medical writer	11.5% (93/809) of articles
Price 2000[22]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	24.1% (40/166) of authors
Mowatt 2002[23]	Individual merited authorship or had assisted with drafting but not listed as an author or acknowledged	8.8% (32/362) of articles
Hao 2009[32]	English-language speakers assisted with writing but not identified as authors or acknowledged	10.4% (NR) of authors
Dotson 2011[28]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	0.9% (1/112) of articles
Wislar 2011[20]	Failure to name, as an author, individuals who made substantial contributions to the research or writing of the article or an unnamed individual who participated in the writing	7.9% (49/622) of articles
Cross-sectional surveys reporting ghost authoring		
Mirzazadeh 2011[24]	Failure to name, as an author, individuals who made substantial contributions to the research	21.4% (25/NR) of authors

Ghajarzadeh 2012[26]	Failure to name, as an author, students who made substantial contributions to the research [c]	0.7% (2/296) of articles
Vinther 2012[21]	Individual merited authorship but not listed as an author	2.4% (6/245) of articles
Rees 2013[31]	Individual merited authorship but not listed as an author	70% (NR/202) of published authors
Publication reviews and descriptive analysis reporting possible ghost authoring or ghostwriting		
Healy 2003[4]	Published articles coordinated by a medical information company, including acknowledged medical writing support [d~~c~~]	57.3% (55/96) of articles
Gøtzsche 2007[30]	Individuals who wrote the trial protocol, conducted the statistical analyses, or wrote the manuscript but were not listed as authors, not members of a study group or steering committee, or not disclosed in an acknowledgment	75.0% (33/44) of trials
Ross 2008[6]	Published reviews associated with Merck support and with a single external author [e~~d~~]	69.4% (50/72) of reviews

[a] Available as online supplementary data.

[b] Values represent the mean weighted percentage of publications that were ghostwritten by respondents. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

[b~~,~~ c] Students were classified as ghostwriters if the student was not named as an author and if the results reported in the publications were based on the results of their theses.

[e~~,~~ d] Authors conclude data provide quantification of the possible extent of ghostwriting based on a single drug. Of the 55 published articles that were coordinated through a medical information company, 2 included medical writing assistance that was acknowledged in the published article.

[d~~,~~ e] Published review articles had been identified from correspondence between Merck and a medical publishing company, from Merck publication status reports, or were affiliated with an author named within the correspondence or publication status reports. The authors did not report whether medical writing assistance was acknowledged in the published article.

Abbreviations: NR, not reported.

questions that required an anonymous response and had response rates greater than 65%. The prevalence of ghost authoring (which included ghostwriting) was estimated to be 11.5% in 1996 and 7.9% in 2008. The prevalence of ghostwriting from these surveys was estimated to be 1.4% in 1996 and 0.2% in 2008. Hamilton and Jacobs conducted a cross-sectional survey of members of two medical writing associations in 2005, 2008, and 2011.[25, 33] The survey, which was not conducted in targeted populations, used pretested questions that required an anonymous response and had response rates from 12% to 28% of the total population surveyed (ie, not all participants surveyed were involved in the preparation of peer-reviewed manuscripts). Findings from these surveys showed that the mean weighted percentage of publications that were ~~association members who contributed to peer-reviewed publications and had~~ ghostwritten by respondents ~~at least once during the survey year~~ had decreased from 61.8% in 2005 to 33.0% in 2011. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

Six cross-sectional survey publications reported the prevalence of possible ghostwriting (ie, the prevalence of ghostwriting and ghost authoring combined).[20, 22, 23, 27, 28, 32] The prevalence of possible ghostwriting reported in these cross-sectional surveys varied from 0.9% to 24.1% of publications or authors.

Four cross-sectional survey publications reported the prevalence of ghost authoring.[21, 24, 26, 31] The prevalence of ghost authoring reported in these cross-sectional surveys varied from 0.7% to 70% of publications or authors.

The descriptive analyses of published articles on sertraline and rofecoxib did not include a prespecified definition of ghost authoring or ghostwriting;[4, 6] in these studies, possible ghostwriting or ghost authoring was assumed in publications associated with industry-sponsored support (Table 2). The descriptive analysis of industry-initiated trials in Denmark used a nonstandard definition of ghost authors; in this study ghost authors were defined as

individuals, not named as authors, who were involved in writing the protocol, conducting the statistical analyses, or writing the manuscript.[30] The prevalence of possible ghostwriting in the two descriptive analyses of single drugs that did not include a prespecified definition of ghostwriting was 57% of articles on sertraline published from 1998 to 2000[4] and 69% of reviews on rofecoxib published from 1996 to 2004.[6] The prevalence of possible ghostwriting in the descriptive analysis of Danish industry-initiated trials, which used a nonstandard definition of authorship, was 75% of Danish initiated trials approved in 1994 to 1995.[30] The descriptive analysis of noninferiority trials[29] was not considered further as the definition of ghostwriting used was consistent with disclosed medical writing assistance. In this study ghostwriters were defined as acknowledged individuals, other than authors, who contributed to the writing and were affiliated with an industry-sponsored study.[29] The prevalence of disclosed medical writing assistance in this descriptive analysis was 17.3% (101/583) of clinical trials.

Secondary publications

Publication characteristics

Of the 32 secondary publications, there were 13 review articles, 10 editorials, 5 commentaries, 3 news articles, and 1 government report, with most being published after 2008 (Table 3). Most publications cited primary sources as evidence of the prevalence of possible ghostwriting (Table 3), with the cited prevalence of possible ghostwriting varying from 6% of publications to 100% of publications involving drugs. In most secondary publications, the information on the prevalence of ghostwriting was not reported consistently compared with the cited evidence (Table 3). Misleading and mistaken information was reported in many publications that (i) did not distinguish between ghostwriting and ghost authoring, (ii) included acknowledged medical writing assistance or a combined estimate of guest authorship and ghost authorship as ghostwriting, (iii) generalized findings from publication reviews and analyses of specific datasets to wider populations of publications or

Table 3. Characteristics of Secondary Publications Citing Evidence of Possible Ghostwriting

Publication	Cited evidence	Consistent with cited source(s)	Comment
Elliot Hastings Cent Rep 2004[35]	Primary	No	Did not distinguish GW from GA
Moffat Perspect Biol Med 2007[36]	Primary	No	Did not distinguish GW from GA
Schiefe Pharmacotherapy 2009[47]	Primary	No	Did not distinguish GW from GA
MacLennan Climacteric 2010[48]	Primary	No	Did not distinguish GW from GA
Nahai Aesthet Surg J 2010[49]	Secondary	No	Did not distinguish GW from GA
Ngai Account Res 2005[50]	Primary	Yes	Did not distinguish GW from GA
Bosch EMBO Rep 2011[43]	Primary	Yes	Did not distinguish GW from GA
Wiwanitkit Am J Med 2012[51]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Krimsky Med Law 2007[52]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Langdon-Neuner Mens Sana Monogr 2008[53]	Primary & secondary	No	Did not distinguish GW from GA or GW from disclosed medical writing support
Tharyan Indian J Med Ethics 2011[45]	Primary	Yes	Did not distinguish GW from GA or GW from disclosed medical writing assistance
Paul Clin Microbiol Infect 2009[54]	Primary	Yes	Did not distinguish GW / GA from disclosed medical writing assistance
Bavdekar Lung India 2012[42]	Primary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Górski Transplant Proc 2010[55]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population

Matias-Guiu Neurologia 2011[44]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
McHenry Mens Sana Monogr 2010[56]	Gov. report	No	Generalized evidence to a wider population
Healy BMJ 2004[57]	Primary & secondary	No / ND	Generalized evidence to a wider population and reported personal opinion of GW prevalence
Abbasi BMJ 2004[2]	Secondary	No	Secondary publication cited named individual
Mitrany Science Editor 2005[58]	None	ND	Cited named individual
Collier Can Med Assoc J 2009[3]	None	ND	Cited named individual
Kmietowicz BMJ 2004[59]	None	ND	Cited named individual
Matthews Wall St J 2005[1]	None	ND	Cited unpublished data
Bonita Heart Fail Clin 2011[60]	Primary	Yes	Consistent with cited source
Grassley Int J Occup Environ Health 2011[61]	Primary	Yes	Consistent with cited source
Nat Clin Pract Nephrol 2007[62]	Primary	Yes	Consistent with cited source
Editors J Urol 2008[15]	Primary	Yes	Consistent with cited source
Baethge Deutsches Arzteblatt 2009[63]	Primary	Yes	Consistent with cited source
Flanagin CSE Annual Mtg 2010[64]	Primary	Yes	Consistent with cited source
Murray Open Med 2010[65]	Primary	Yes	Consistent with cited source
Moore BMJ 2004[66]	Secondary	Yes	Consistent with cited source
Hargreaves BMJ 2007[67]	Primary	Yes	Consistent with cited source
Jones Nature 2009[68]	Primary	Yes	Consistent with cited source

Abbreviations: CSE, Council of Science Editors; GA, ghost authoring; Gov, government; GW, ghostwriting; ND, not determined.

industry-sponsored trials, or (iv) cited personal communications or informal surveys where the original source was unpublished and could not be verified (Table 3).

Case study of misleading and mistaken evidence

An analysis of secondary publications citing the possible prevalence of ghostwriting in sertraline publications provides an illuminating case study on how misleading or mistaken evidence can enter and remain in the medical literature. In 2003, Healy and Cattell showed that 57% (55/96) of articles on sertraline published from 1998 to 2000 were coordinated through a medical information company and only two acknowledged medical writing assistance.[4] The authors concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". In 2004 (October 14), Healy also provided evidence on the influence of the pharmaceutical industry on key groups at a United Kingdom House of Commons Health Select Committee investigation.[34] In answer to Question 197, Healy stated the following:

"My estimate is that, even in journals like the BMJ, the Lancet, the New England Journal of Medicine and JAMA, the leading journals in the field, if these articles have to do with therapeutics, with drugs, it may be worse perhaps for psychiatry than elsewhere, but I doubt it, 50% of these articles are ghostwritten. It may be higher."

Healy and Cattell's original evidence has been cited incorrectly and interchangeably with Healy's statement to the House of Commons Health Select Committee investigation (Figure 2). Although findings from the primary publication have been cited and interpreted correctly in two secondary publications,[35, 36] many secondary publications incorrectly cited Healy's original evidence and statement to the House of Commons Health Select Committee investigation (Figure 2).

DISCUSSION

This systematic review on the reported prevalence of ghostwriting in the medical literature showed that the estimates of the prevalence of ghostwriting in primary publications varied markedly and were influenced by the definitions used, the types of study designs, and the type of population or samples assessed. In addition, secondary publications often cited outdated, misleading, or mistaken evidence of the reported prevalence of ghostwriting, with many publications not distinguishing ghostwriting from ghost authoring.

Although evidence from descriptive analyses can highlight the extent of ghostwriting in single populations, evidence from well-conducted cross-sectional surveys have the potential to provide estimates of the prevalence of ghostwriting that may be generalized to the majority of peer-reviewed publications. Despite this, many of the cross-sectional surveys retrieved in this systematic review were conducted in limited populations that were not broadly representative of the peer-reviewed medical literature. In addition, many of the cross-sectional surveys did not differentiate between contributions that merited authorship from those that did not merit authorship and provided at best, an estimate of possible ghostwriting. The reported prevalence of ghostwriting, where ghostwriting was defined as undisclosed contributions that did not merit authorship, was retrieved from the two cross-sectional surveys of corresponding authors from several general medicine journals and by cross-sectional surveys (repeated on three separate occasions from 2005 to 2011) of members of two major medical writing associations. Together the findings from these surveys suggested that the prevalence of ghostwriting has decreased in recent years. However, while the findings from these surveys may be considered more broadly representative of the peer-reviewed medical literature than surveys focused on single journals, single countries, or single subject areas, interpretation of these findings should take

into account that respondents were required to retrospectively self-report potentially unethical or unprofessional behaviour.

Unethical authorship practices are a major concern and are an increasingly recognized problem in the medical literature.[37] As the findings from this systematic review suggest, some of these perceived problems may arise from the considerable, but unnecessary, confusion and disagreement surrounding the definitions of ghostwriting. As recognized by the World Association of Medical Editors, professional medical writers can have a legitimate role in assisting authors to communicate their research findings in the peer-reviewed literature.[14] Professional medical writers can and do improve the timeliness and quality of reporting and can assist investigators and industry sponsors to meet their ethical commitments to the disclosure and publishing of clinical trial results.[7, 38-40] As such, the misleading and mistaken reporting of the prevalence of ghostwriting that was evident in the secondary publications retrieved in this systematic review is disappointing. Specifically, the confusion surrounding definitions of ghost authorship and ghostwriting and the unbalanced focus on industry as the source of unethical authorship practices in the secondary publications takes attention away from the need to focus on all types of unethical contributions in peer-reviewed publications, irrespective of the source of the unethical practice.

The strengths of this systematic review are that a broad search strategy was employed with few restrictions to minimize any potential for publication or language bias. All study designs and publication types were considered and there were no restrictions on language. Although the full text or abstracts of 8 publications could not be retrieved, omission of these publications was unlikely to have biased the findings. Review of the abstracts or titles of these publications suggested that none reported numerical estimates of the prevalence of ghostwriting. The major finding of this review was the limitations of the reported evidence of ghostwriting in the medical literature. These limitations included the heterogeneity among the

studies in the outcomes reported and populations investigated, the observational study designs, and the retrospective nature of data collection. Given the nature of unethical authorship practices, it may not be feasible to conduct a prospective study on ghostwriting. However, based on the findings from this review, recommendations can be made to help researchers, authors, editors, and peer-reviewers apply the same rigorous standards that are applied to the conduct and assessment of all clinical research and actively improve the quality of reporting of the evidence of unethical authorship practices.

Researchers should use a standard definition of ghostwriting so that the confusion around ghostwriters and ghost authors is not perpetuated. Ghostwriting is paid or unpaid writing assistance by individuals who generally do not merit authorship and whose contributions are not disclosed in the acknowledgments.[7] In contrast, ghost authoring is contributions to the research, data analysis, and/or writing of a manuscript by individuals who do merit authorship and whose contributions are not disclosed in the author byline.[7] For example, the descriptive analysis of Danish industry-initiated trials used a nonstandard definition of authorship, which is likely to have contributed to the very high prevalence of possible ghostwriting reported in this study (75%). In this study, Gøtzsche et al.[30] suggested that individuals who write the trial protocol, conduct the statistical analyses, or who contribute to the writing of a publication should be included as authors. Indeed, the prevalence of ghost authorship in this study was 91% when the analyses included these individuals, irrespective of whether they had been appropriately acknowledged elsewhere. Given that the Gøtzsche et al.[30] study has been downloaded more than 3,000 times since publication[41] and cited repeatedly as evidence not only of ghost authorship but also of ghostwriting,[42-45] it would have been illuminating if the authors had included an estimate of the prevalence of ghostwriting using standard definitions for comparison. In addition to using standard definitions of ghostwriting, researchers and authors should ensure that cited evidence of ghostwriting is reported accurately without unwarranted generalizations. Publications based on personal commentary should be avoided and studies that use nonstandard definitions,

specific populations, or that were conducted before a change in practice (eg, before the adoption of the Good Publication Practice guidelines for communicating company sponsored research in 2003 [GPP][46] and in 2009 [GPP2][11]) should be described in context.

Editors and peer reviewers have a duty to prevent outdated, misleading, or mistaken evidence on the prevalence of ghostwriting from being published and perpetuated. Close attention should be paid to the internal and external validity of the study, the definitions used, how the data are reported, and whether the data are interpreted within the context of current practices. When assessing a submitted manuscript on ghostwriting, editors should consider using peer reviewers with expertise in the appropriate study designs (eg, survey research) and ethical publication practices. Collectively, these actions could help prevent further questionable evidence on the prevalence of ghostwriting from being published.

In conclusion, the findings from this systematic review showed that reports of the prevalence of ghostwriting in the medical literature are limited by the varied definitions used to describe unethical authorship practices, the types of study designs employed, and the populations assessed. To improve reporting, researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. While open and transparent debate should be encouraged, editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

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COMPETING INTERESTS

The author is a professional medical writer who is actively involved in national and international not-for-profit organizations that encourage ethical medical writing practices. The author has no relationships with any pharmaceutical company that might have an interest in the submitted work and has no other relationships that could be construed as a competing interest.

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FIGURE LEGENDS

Figure 1. Selection of publications reporting a numerical estimate of the prevalence of ghostwriting. Electronic databases were searched on 23 May 2013.

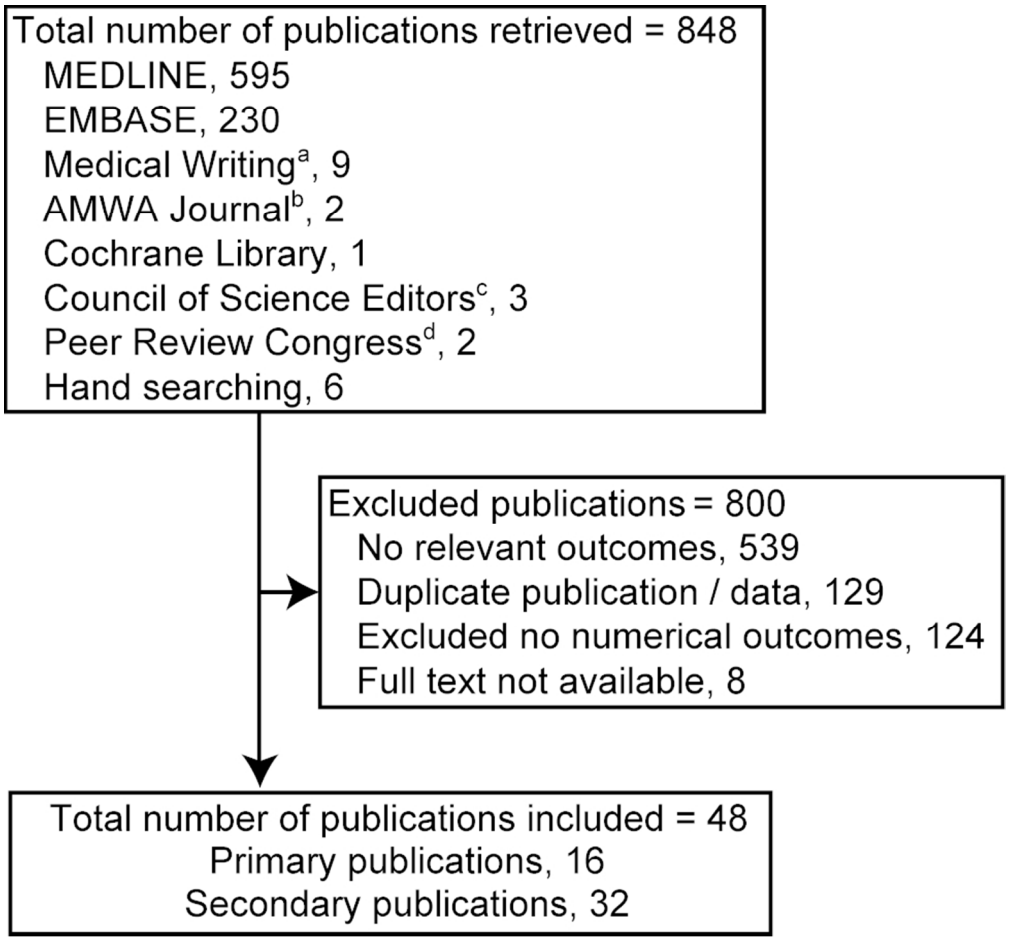
[a] Journal of the European Medical Writers Association, formerly known as The Write Stuff.

[b] Journal of the American Medical Writers Association.

[c] Council of Science Editors Annual Meetings.

[d] International Congress on Peer Review and Biomedical Publication.

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[4] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.



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a Journal of the European Medical Writers Association, formerly known as The Write Stuff.
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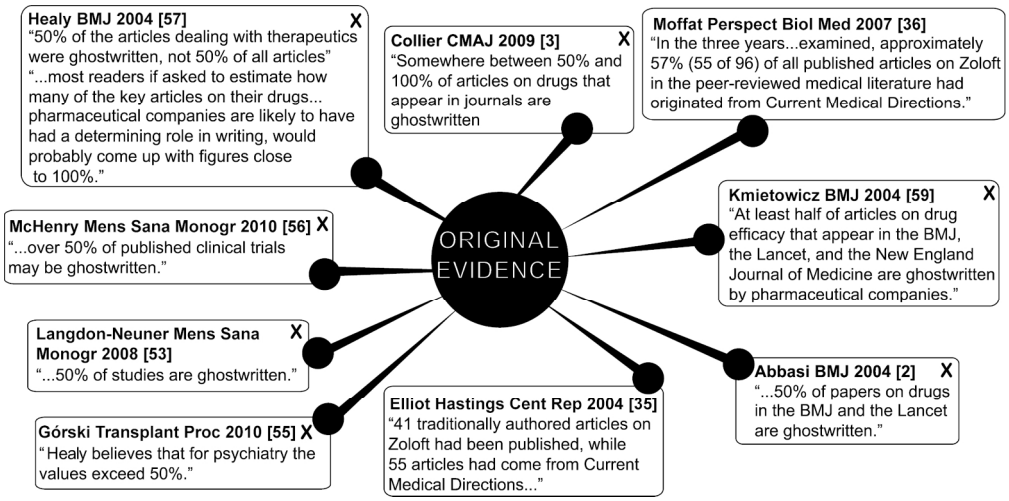
82x76mm (300 x 300 DPI)



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170x83mm (300 x 300 DPI)

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	p. 1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	pp. 2/3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	p. 5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	p. 6
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	A review protocol was prepared that prespecified the primary and secondary objectives of the literature review, eligibility criteria, data to be collected, and primary and secondary outcome measures. The review protocol was not registered in a public registry but is available upon request.
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	p. 7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	p. 6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	p. 6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	pp. 6/7
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	p. 7

PRISMA 2009 Checklist

Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	p. 7
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	Risk of bias was not conducted formally because data were not synthesised and because of the various study types retrieved. Given the various study designs and methods used across the retrieved studies, the quality of the study methods were assessed on a study-by-study basis. The key factors considered were the study design, definition of ghostwriting used, and study population. The factors considered when determining the quality of the cross-sectional surveys are described in the Methods section (p. 7).
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	Not applicable because study data were not synthesised
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	Not applicable because study data were not synthesised.

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	Risk of bias across studies was not conducted formally because data were not synthesised and because of the considerable variation in study designs, populations, and definitions of ghostwriting used in the retrieved publications.
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	No additional analyses were conducted
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	8, Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	8-11, 15, Tables 1-3
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Risk of bias was not conducted formally because data were not synthesised. All studies retrieved were limited by the observational nature of the data collection. The quality of individual components of each study are listed in Table 1

PRISMA 2009 Checklist

			and are summarised in the results section (Publication Characteristics) (pp. 8/9).
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Tables 1-3
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	Not applicable because study data were not synthesised
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	Not applicable because study data were not synthesised and because of the considerable variation in study designs, populations, and definitions of ghostwriting used in the retrieved publications.
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	No additional analyses were conducted
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	20-24
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	20-22
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	22-24
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	No funding was received to conduct this study

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: **www.prisma-statement.org**.

BMJ Open

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

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Secondary Subject Heading:	Ethics
Keywords:	MEDICAL ETHICS, systematic review, ghostwriting, authorship

TITLE

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

AUTHOR

Serina Stretton, PhD CMPP

AUTHOR'S AFFILIATION

ProScribe – Envision Pharma Group, Sydney, NSW, Australia

AUTHOR FOR CORRESPONDENCE

Serina Stretton, PhD, CMPP

ProScribe – Envision Pharma Group

18 Poulet St

Matraville, NSW, 2036

AUSTRALIA

Tel: + 612 9661 0768

Fax: + 617 5474 9156

Email: serina.stretton@EnvisionPharmaGroup.com

KEYWORDS

Authorship, ghostwriting, systematic review

WORD COUNT

3,967

ABSTRACT

Background: Ghostwriting of industry-sponsored articles is unethical and is perceived to be common practice.

Objective: To systematically review how evidence for the prevalence of ghostwriting is reported in the medical literature.

Data sources: MEDLINE via PubMed 1966+, EMBASE 1966+, The Cochrane Library 1988+, Medical Writing 1998+, The AMWA Journal 1986+, Council of Science Editors Annual Meetings 2007+, and the Peer Review Congress 1994+ were searched electronically (23 May 2013) using the search terms ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*.

Eligibility criteria: All publication types were considered; only publications reporting a numerical estimate of possible ghostwriting prevalence were included.

Data extraction: Two independent reviewers screened the publications; discrepancies were resolved by consensus. Data to be collected included a numerical estimate of the prevalence of possible ghostwriting (primary outcome measure), definitions of ghostwriting reported, source of the reported prevalence, publication type and year, study design, and sample population.

Results: Of the 848 publications retrieved and screened for eligibility, 48 reported numerical estimates for the prevalence of possible ghostwriting. Sixteen primary publications reported findings from cross-sectional surveys or descriptive analyses of published articles; 32 secondary publications cited published or unpublished evidence. Estimates on the prevalence of possible ghostwriting in primary and secondary publications varied markedly. Primary estimates were not suitable for meta-analysis because of the various definitions of ghostwriting used, study designs, and types of populations or samples. Secondary estimates were not always reported or cited correctly or appropriately.

Conclusions: Evidence for the prevalence of ghostwriting in the medical literature is limited and can be outdated, misleading, or mistaken. Researchers should not inflate estimates

using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. Editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

Strengths

- First systematic review on the reporting of the prevalence of ghostwriting in the medical literature
- A broad search strategy was employed with few restrictions to minimize any potential for publication or language bias; there were no restrictions on language.
- All study designs and publication types were considered

Limitations

- Study heterogeneity in the outcomes reported and populations investigated precluded synthesis of the data
- Retrospective and self-reported nature of the data collection increased the risk of selection bias in the studies
- Most included studies were not broadly representative of the peer-reviewed medical literature

INTRODUCTION

Ghostwriting occurs when paid or unpaid writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments.[1] This practice is considered to be distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[1] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding these unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[2]

Ghostwriting of peer-reviewed journal publications in the medical literature is believed to be common practice.[3-5] This belief is supported, to a large extent, by highly publicized cases, primarily from the 1990s and early 2000s, of pharmaceutical companies and authors who had used ghostwriters to prepare manuscripts for publication in medical journals.[6-8] Such cases are highly unethical because the role of the commercial sponsor and any other potential conflicts of interest were hidden. Estimating the prevalence of ghostwriting in the medical literature has been hindered not only by the different definitions of ghostwriting and ghost authoring, but also by the failure of authors to distinguish ghostwriting, which is unethical, from professional medical writing support, which is ethical.[9, 10] Ghostwriters keep their involvement in a manuscript hidden, whereas professional medical writers disclose their involvement and follow ethical publication practices.[1, 9-11] Consistent with the authorship criteria recommended by the International Committee of Medical Journal Editors (ICMJE), professional medical writers who provide writing assistance and do not meet all of the ICMJE authorship criteria, should be acknowledged rather than listed as authors.[12] The prevalence of disclosed professional medical writing assistance in medical journals has been estimated to be between 6.0% and 11.0%[10, 13] and the legitimate value

that this medical writing assistance can bring to improving the quality, timeliness, and integrity of reporting in medical journals has been demonstrated.[1, 14, 15] However, the exact prevalence of ghostwriting and other forms of undisclosed contributions to papers published in medical journals is unknown.

The primary objective of this study was to conduct the first systematic review on how the prevalence of ghostwriting is reported in the medical literature. The secondary objectives were to assess the variability of the reported estimates of the prevalence of ghostwriting and investigate the source for these estimates.

MATERIALS AND METHODS

Literature search strategy

A search strategy was developed to retrieve publications from the medical literature that reported quantitative estimates of the prevalence of ghostwriting. The following databases were searched on 23 May 2013: MEDLINE via PubMed (1966+); EMBASE (1966+); The Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials (1988+); The AMWA Journal (1986+, journal of the American Medical Writers Association); Medical Writing (1998+, journal of the European Medical Writers Association, formerly known as The Write Stuff); Council of Science Editors annual meetings (2007+); and the International Congress on Peer Review and Biomedical Publication meetings (1994+). General text was searched using the following search terms: ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*. Truncation symbols and Boolean operators (AND, OR) were used wherever possible.

Two reviewers (SS, SMG) independently screened the title and abstracts of all retrieved publications using prespecified eligibility criteria; the full text of potentially eligible publications was screened to confirm eligibility for inclusion. Any discrepancies between the reviewers were resolved by consensus. The reference lists of relevant reviews and other publications were screened by hand to identify any additional publications for inclusion.

Eligibility criteria

The eligibility criteria were prespecified. Publications were included if they reported a numerical estimate of the prevalence of possible ghostwriting. Publications were excluded if they were duplicate publications from different databases or abstracts of subsequently published full-text articles, did not report any outcomes related to ghostwriting, or reported qualitative estimate(s) of the prevalence of possible ghostwriting. No restrictions on language were included in the eligibility criteria. If needed, the abstracts of publications

written in non-English language were to be translated to assist in screening for eligibility. If publications were eligible for inclusion, the full text of the publications were to be translated to English.

Data extraction and analysis

The primary and secondary outcomes and data to be collected were prespecified. Data to be collected included publication type and year, study design and sample population, definitions of ghostwriting reported, reported prevalence of possible ghostwriting, and the source of the reported prevalence of ghostwriting. The prevalence of possible ghostwriting was reported as published. No unpublished data from the retrieved literature were reported and values for prevalence were not extrapolated from published data.

The primary outcome measure for the systematic review was a numerical estimate of the prevalence of possible ghostwriting. The reported definitions of possible ghostwriting were categorized according to the following standard definitions, irrespective of the term used to describe the practice in the publication. Undisclosed writing contributions to a manuscript were defined as (i) ghostwriting if they were described as not meriting authorship and were not listed in the acknowledgments[9, 16] and (ii) ghost authoring if they did merit authorship and were not listed in the author byline.[1] Consistent with recommendations from international medical journal editors,[12, 14] Good Publication Practice guidelines,[11] and professional medical writing associations,[17, 18] disclosed writing contributions to a manuscript that did not merit authorship and were disclosed in the acknowledgments were not categorized as ghostwriting.

The key factors considered when assessing study quality were study design, the population assessed, and how ghostwriting was defined. While there is no consensus on the best practice for reporting survey research,[19] the quality of the cross-sectional surveys was

determined by an assessment of the following factors: validation or pretesting of the survey questions, anonymity of the response, sample size, and response rate.

RESULTS

Publication selection

A total of 848 articles were retrieved from the literature search, 800 were excluded, and 48 met the eligibility criteria and were included in the systematic review (Figure 1). The main reasons for exclusion were publications not relevant to ghostwriting (n = 539), duplicate publications from different databases (n = 129), and publications not reporting a numerical estimate of the prevalence of possible ghostwriting (n = 124). The titles or abstracts of the 8 publications that were excluded because the full text or abstract could not be retrieved were reviewed; none were considered to report a numerical estimate of the prevalence of possible ghostwriting. Overall, eligible publications included 16 primary publications that reported original research on the prevalence of possible ghostwriting and 32 secondary publications that cited published or unpublished evidence of possible ghostwriting.

Primary publications

Publication characteristics

Of the 16 primary publications (Table 1), there were 13 full-text publications[6, 8, 20-30] and three conference abstracts[31-33] that reported a numerical estimate of possible ghostwriting. Twelve publications[20-28, 31-33] reported findings from cross-sectional surveys and four publications[6, 8, 29, 30] reported findings from descriptive analyses of published articles. Most cross-sectional surveys were reasonably well reported. The surveys used were not validated but most included pretested questions, required an anonymous response, were conducted in targeted populations (ie, all individuals invited to participate were involved in the preparation of peer-reviewed manuscripts), and reported a sample size and response rate. Of the cross-sectional surveys conducted in targeted populations (eg, corresponding or first authors), six had response rates greater than 50%, two had response rates less than 50%, and one did not report a response rate (Table 1). Of the cross-sectional surveys conducted in nontargeted populations (eg, individuals invited to participate who may

or may not have been involved in providing medical writing assistance for peer-reviewed publications), three had low (12% to 28%) response rates and one did not report a response rate (Table 1).

Table 1. Characteristics of Primary Publications Reporting Original Evidence of Ghostwriting

Publication	Population	Survey description	Data collected	N	RR
Cross-sectional surveys – authors or corresponding authors					
Flanagin 1998[27]	6 general medicine journals [a]	Self-administered, postal, anonymous, pretested, targeted	1996	809	69%
Mowatt 2002[23]	Published Cochrane reviews	Self-administered, online, anonymous, pretested, targeted	1999	362	63%
Hao 2009[32] (Abstract)	Chin Med J	Self-administered, email, not anonymous, targeted	2008 [c]	220	86%
Dotson 2011[28]	3 pharmacy journals	Self-administered, online, anonymous, pretested, targeted	2009	112	25%
Mirzazadeh 2011[24]	3 Iranian journals	Self-administered, email, not anonymous, targeted	2009-2010	NR	NR
Wislar 2011[20]	6 general medicine journals [b]	Self-administered, online, anonymous, pretested, targeted	2008	622	70%
Ghajarzadeh 2012[26]	Arch Iran Med (based on student theses)	Self-administered, email, anonymous, targeted	2005-2007	30	49%
Vinther 2012[21]	Ugeskr Laeger & Dan Med J	Self-administered, online, anonymous, pretested, targeted	2010	272	62%
Cross-sectional surveys – healthcare professionals					
Price 2000[22]	Health academic staff	Self-administered, postal, anonymous, pretested, targeted	NR	166	59%
Rees 2013[31] (Abstract)	Registered users of EPG Online	Self-administered, online, anonymous, nontargeted	NR	295	NR

Cross-sectional surveys – members of medical writing associations					
Jacobs 2009[25]	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2005 2008	843 773	28% 14%
Hamilton 2012[33] (Abstract)	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2011	620	12% [d]
Publication reviews and descriptive analysis					
Healy 2003[6]	Articles on sertraline	NA	1998-2000	96	NA
Gøtzsche 2007[30]	Articles on Danish industry-initiated trials approved 1994-1995	NA	NR	44	NA
Ross 2008[8]	Reviews on rofecoxib associated with Merck support	NA	1996-2004	72	NA
Suda 2011[29]	Noninferiority clinical trials	NA	1989-2009	583	NA

[a] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, American Journal of Cardiology, American Journal of Medicine, American Journal of Obstetrics and Gynecology.

[b] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, Lancet, Nature Medicine, PLoS Medicine.

[c] Submission date.

[d] Personal communication, C. Hamilton.

Abbreviations: AMWA, American Medical Writers Association; EMWA, European Medical Writers Association; NA, not applicable; NR, not reported; RR, response rate.

Not all cross-sectional surveys were broadly representative of the peer-reviewed medical literature. The cross-sectional surveys were conducted in single populations of academic staff or healthcare professionals[22, 31], members of medical writing associations[25, 33], or corresponding or first authors. The corresponding or first authors were surveyed from single journals (Cochrane reviews, Chinese Medical Journal, Archives of Iranian Medicine),[23, 26, 32] two Danish journals,[21] three Iranian journals,[24] and three pharmacy journals.[28] Two cross-sectional surveys of corresponding authors were conducted in six general medicine journals.[20, 27]

The descriptive analyses of published articles were conducted in single populations and included an analysis of sertraline publications from 1998 to 2000,[6] rofecoxib reviews from 1996 to 2004,[8] publications from industry-initiated trials in Denmark from 1994 to 1995,[30] and from published noninferiority trials from 1989 to 2009.[29]

Reported prevalence of ghostwriting

The reported prevalence of possible ghostwriting in the primary publications varied markedly and were not suitable for meta-analysis because of the different populations assessed, the different methods used to generate the estimates, and the various definitions that were used (Tables 1 and 2). All cross-sectional surveys reported a definition of ghostwriting or ghost authoring, but most definitions did not differentiate contributions that merited authorship from those that did not merit authorship (Table 2).

Table 2. Primary Publications Reporting Estimates of the Prevalence of Possible Ghostwriting

Publication	Measure of Possible Ghostwriting Reported by Authors	Estimate % (n)
Cross-sectional surveys reporting ghostwriting		
Flanagin 1998[27]	Unnamed individual who participated in the writing	1.4% (11/809) of articles
Wislar 2011[20]	Unnamed individual who participated in the writing	0.2% [a] (1/622) of articles
Jacobs 2009, Hamilton 2012[25, 33]	Undisclosed medical writing assistance not qualifying for authorship	2005: 61.8% (NR) 2009: 41.7% (NR) 2011: 33.0% (NR) of articles [b]
Cross-sectional surveys reporting combined ghost authoring and ghostwriting		
Flanagin 1998[27]	Failure to name, as an author, individuals who made substantial contributions to the research or writing or an unidentified medical writer	11.5% (93/809) of articles
Price 2000[22]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	24.1% (40/166) of authors
Mowatt 2002[23]	Individual merited authorship or had assisted with drafting but not listed as an author or acknowledged	8.8% (32/362) of articles
Hao 2009[32]	English-language speakers assisted with writing but not identified as authors or acknowledged	10.4% (NR) of authors
Dotson 2011[28]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	0.9% (1/112) of articles
Wislar 2011[20]	Failure to name, as an author, individuals who made substantial contributions to the research or writing of the article or an unnamed individual who participated in the writing	7.9% (49/622) of articles
Cross-sectional surveys reporting ghost authoring		
Mirzazadeh 2011[24]	Failure to name, as an author, individuals who made substantial contributions to the research	21.4% (25/NR) of authors
Ghajarzadeh	Failure to name, as an author, students who made substantial	0.7% (2/296) of

2012[26]	contributions to the research [c]	articles
Vinther 2012[21]	Individual merited authorship but not listed as an author	2.4% (6/245) of articles
Rees 2013[31]	Individual merited authorship but not listed as an author	70% (NR/202) of published authors
Publication reviews and descriptive analysis reporting possible ghost authoring or ghostwriting		
Healy 2003[6]	Published articles coordinated by a medical information company, including acknowledged medical writing support [d]	57.3% (55/96) of articles
Gøtzsche 2007[30]	Individuals who wrote the trial protocol, conducted the statistical analyses, or wrote the manuscript but were not listed as authors, not members of a study group or steering committee, or not disclosed in an acknowledgment	75.0% (33/44) of trials
Ross 2008[8]	Published reviews associated with Merck support and with a single external author [e]	69.4% (50/72) of reviews

[a] Available as online supplementary data.

[b] Values represent the mean weighted percentage of publications that were ghostwritten by respondents. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

[c] Students were classified as ghostwriters if the student was not named as an author and if the results reported in the publications were based on the results of their theses.

[d] Authors conclude data provide quantification of the possible extent of ghostwriting based on a single drug. Of the 55 published articles that were coordinated through a medical information company, 2 included medical writing assistance that was acknowledged in the published article.

[e] Published review articles had been identified from correspondence between Merck and a medical publishing company, from Merck publication status reports, or were affiliated with an author named within the correspondence or publication status reports. The authors did not report whether medical writing assistance was acknowledged in the published article.

Abbreviations: NR, not reported.

Four cross-sectional surveys reported the prevalence of ghostwriting where the definition provided could be categorized as undisclosed contributions that do not merit authorship.[20, 25, 27, 33] Findings from these surveys, which were repeated at different time points, suggested that the prevalence of ghostwriting is low and decreasing. Flanagin[27] and Wislar[20] conducted two similar cross-sectional surveys of corresponding authors of articles published in six general medicine journals in 1996[27] and in 2008.[20] The surveys, which included a core set of three journals (Annals of Internal Medicine, JAMA, the New England Journal of Medicine) at each time point and in targeted populations, used pretested questions that required an anonymous response and had response rates greater than 65%. The prevalence of ghost authoring (which included ghostwriting) was estimated to be 11.5% in 1996 and 7.9% in 2008. The prevalence of ghostwriting from these surveys was estimated to be 1.4% in 1996 and 0.2% in 2008. Hamilton and Jacobs conducted a cross-sectional survey of members of two medical writing associations in 2005, 2008, and 2011.[25, 33] The survey, which was not conducted in targeted populations, used pretested questions that required an anonymous response and had response rates from 12% to 28% of the total population surveyed (ie, not all participants surveyed were involved in the preparation of peer-reviewed manuscripts). Findings from these surveys showed that the mean weighted percentage of publications that were ghostwritten by respondents had decreased from 61.8% in 2005 to 33.0% in 2011. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

Six cross-sectional survey publications reported the prevalence of possible ghostwriting (ie, the prevalence of ghostwriting and ghost authoring combined).[20, 22, 23, 27, 28, 32] The prevalence of possible ghostwriting reported in these cross-sectional surveys varied from 0.9% to 24.1% of publications or authors.

Four cross-sectional survey publications reported the prevalence of ghost authoring.[21, 24, 26, 31] The prevalence of ghost authoring reported in these cross-sectional surveys varied from 0.7% to 70% of publications or authors.

The descriptive analyses of published articles on sertraline and rofecoxib did not include a prespecified definition of ghost authoring or ghostwriting;[6, 8] in these studies, possible ghostwriting or ghost authoring was assumed in publications associated with industry-sponsored support (Table 2). The descriptive analysis of industry-initiated trials in Denmark used a nonstandard definition of ghost authors; in this study ghost authors were defined as individuals, not named as authors, who were involved in writing the protocol, conducting the statistical analyses, or writing the manuscript.[30] The prevalence of possible ghostwriting in the two descriptive analyses of single drugs that did not include a prespecified definition of ghostwriting was 57% of articles on sertraline published from 1998 to 2000[6] and 69% of reviews on rofecoxib published from 1996 to 2004.[8] The prevalence of possible ghostwriting in the descriptive analysis of Danish industry-initiated trials, which used a nonstandard definition of authorship, was 75% of Danish initiated trials approved in 1994 to 1995.[30] The descriptive analysis of noninferiority trials[29] was not considered further as the definition of ghostwriting used was consistent with disclosed medical writing assistance. In this study ghostwriters were defined as acknowledged individuals, other than authors, who contributed to the writing and were affiliated with an industry-sponsored study.[29] The prevalence of disclosed medical writing assistance in this descriptive analysis was 17.3% (101/583) of clinical trials.

Secondary publications

Publication characteristics

Of the 32 secondary publications, there were 13 review articles, 10 editorials, 5 commentaries, 3 news articles, and 1 government report, with most being published after 2008 (Table 3). Most publications cited primary sources as evidence of the prevalence of

possible ghostwriting (Table 3), with the cited prevalence of possible ghostwriting varying from 6% of publications to 100% of publications involving drugs. In most secondary publications, the information on the prevalence of ghostwriting was not reported consistently compared with the cited evidence (Table 3). Misleading and mistaken information was reported in many publications that (i) did not distinguish between ghostwriting and ghost authoring, (ii) included acknowledged medical writing assistance or a combined estimate of guest authorship and ghost authorship as ghostwriting, (iii) generalized findings from publication reviews and analyses of specific datasets to wider populations of publications or industry-sponsored trials, or (iv) cited personal communications or informal surveys where the original source was unpublished and could not be verified (Table 3).

Table 3. Characteristics of Secondary Publications Citing Evidence of Possible Ghostwriting

Publication	Cited evidence	Consistent with cited source(s)	Comment
Elliot Hastings Cent Rep 2004[34]	Primary	No	Did not distinguish GW from GA
Moffat Perspect Biol Med 2007[35]	Primary	No	Did not distinguish GW from GA
Schiefe Pharmacotherapy 2009[36]	Primary	No	Did not distinguish GW from GA
MacLennan Climacteric 2010[37]	Primary	No	Did not distinguish GW from GA
Nahai Aesthet Surg J 2010[38]	Secondary	No	Did not distinguish GW from GA
Ngai Account Res 2005[39]	Primary	Yes	Did not distinguish GW from GA
Bosch EMBO Rep 2011[40]	Primary	Yes	Did not distinguish GW from GA
Wiwanitkit Am J Med 2012[41]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Krimsky Med Law 2007[42]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Langdon-Neuner Mens Sana Monogr 2008[43]	Primary & secondary	No	Did not distinguish GW from GA or GW from disclosed medical writing support

Tharyan Indian J Med Ethics 2011[44]	Primary	Yes	Did not distinguish GW from GA or GW from disclosed medical writing assistance
Paul Clin Microbiol Infect 2009[45]	Primary	Yes	Did not distinguish GW / GA from disclosed medical writing assistance
Bavdekar Lung India 2012[46]	Primary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Górski Transplant Proc 2010[47]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Matias-Guiu Neurologia 2011[48]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
McHenry Mens Sana Monogr 2010[49]	Gov. report	No	Generalized evidence to a wider population
Healy BMJ 2004[50]	Primary & secondary	No / ND	Generalized evidence to a wider population and reported personal opinion of GW prevalence
Abbasi BMJ 2004[4]	Secondary	No	Secondary publication cited named individual
Mitrany Science Editor 2005[51]	None	ND	Cited named individual
Collier Can Med Assoc J 2009[5]	None	ND	Cited named individual
Kmietowicz BMJ 2004[52]	None	ND	Cited named individual
Matthews Wall St J 2005[3]	None	ND	Cited unpublished data
Bonita Heart Fail Clin 2011[53]	Primary	Yes	Consistent with cited source
Grassley Int J Occup Environ Health 2011[54]	Primary	Yes	Consistent with cited source

Nat Clin Pract Nephrol 2007[55]	Primary	Yes	Consistent with cited source
Editors J Urol 2008[15]	Primary	Yes	Consistent with cited source
Baethge Deutsches Arzteblatt 2009[56]	Primary	Yes	Consistent with cited source
Flanagin CSE Annual Mtg 2010[57]	Primary	Yes	Consistent with cited source
Murray Open Med 2010[58]	Primary	Yes	Consistent with cited source
Moore BMJ 2004[59]	Secondary	Yes	Consistent with cited source
Hargreaves BMJ 2007[60]	Primary	Yes	Consistent with cited source
Jones Nature 2009[61]	Primary	Yes	Consistent with cited source

Abbreviations: CSE, Council of Science Editors; GA, ghost authoring; Gov, government; GW, ghostwriting; ND, not determined.

Case study of misleading and mistaken evidence

An analysis of secondary publications citing the possible prevalence of ghostwriting in sertraline publications provides an illuminating case study on how misleading or mistaken evidence can enter and remain in the medical literature. In 2003, Healy and Cattell showed that 57% (55/96) of articles on sertraline published from 1998 to 2000 were coordinated through a medical information company and only two acknowledged medical writing assistance.[6] The authors concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". In 2004 (October 14), Healy also provided evidence on the influence of the pharmaceutical industry on key groups at a United Kingdom House of Commons Health Select Committee investigation.[62] In answer to Question 197, Healy stated the following:

"My estimate is that, even in journals like the BMJ, the Lancet, the New England Journal of Medicine and JAMA, the leading journals in the field, if these articles have to do with therapeutics, with drugs, it may be worse perhaps for psychiatry than elsewhere, but I doubt it, 50% of these articles are ghostwritten. It may be higher."

Healy and Cattell's original evidence has been cited incorrectly and interchangeably with Healy's statement to the House of Commons Health Select Committee investigation (Figure 2). Although findings from the primary publication have been cited and interpreted correctly in two secondary publications,[34, 35] many secondary publications incorrectly cited Healy's original evidence and statement to the House of Commons Health Select Committee investigation (Figure 2).

DISCUSSION

This systematic review on the reported prevalence of ghostwriting in the medical literature showed that the estimates of the prevalence of ghostwriting in primary publications varied markedly and were influenced by the definitions used, the types of study designs, and the type of population or sample assessed. In addition, secondary publications often cited outdated, misleading, or mistaken evidence of the reported prevalence of ghostwriting, with many publications not distinguishing ghostwriting from ghost authoring.

Although evidence from descriptive analyses can highlight the extent of ghostwriting in single populations, evidence from well-conducted cross-sectional surveys have the potential to provide estimates of the prevalence of ghostwriting that may be generalized to the majority of peer-reviewed publications. Despite this, many of the cross-sectional surveys retrieved in this systematic review were conducted in limited populations that were not broadly representative of the peer-reviewed medical literature. In addition, many of the cross-sectional surveys did not differentiate between contributions that merited authorship from those that did not merit authorship and provided at best, an estimate of possible ghostwriting. The reported prevalence of ghostwriting, where ghostwriting was defined as undisclosed contributions that did not merit authorship, was retrieved from the two cross-sectional surveys of corresponding authors from several general medicine journals and by cross-sectional surveys (repeated on three separate occasions from 2005 to 2011) of members of two major medical writing associations. Together the findings from these surveys suggested that the prevalence of ghostwriting has decreased in recent years. However, while the findings from these surveys may be considered more broadly representative of the peer-reviewed medical literature than surveys focused on single journals, single countries, or single subject areas, interpretation of these findings should take into account that respondents were required to retrospectively self-report potentially unethical or unprofessional behaviour.

Unethical authorship practices are a major concern and are an increasingly recognized problem in the medical literature.[63] As the findings from this systematic review suggest, some of these perceived problems may arise from the considerable, but unnecessary, confusion and disagreement surrounding the definitions of ghostwriting. As recognized by the World Association of Medical Editors, professional medical writers can have a legitimate role in assisting authors to communicate their research findings in the peer-reviewed literature.[14] Professional medical writers can and do improve the timeliness and quality of reporting and can assist investigators and industry sponsors to meet their ethical commitments to the disclosure and publishing of clinical trial results.[1, 64-66] As such, the misleading and mistaken reporting of the prevalence of ghostwriting that was evident in the secondary publications retrieved in this systematic review is disappointing. Specifically, the confusion surrounding definitions of ghost authorship and ghostwriting and the unbalanced focus on industry as the source of unethical authorship practices in the secondary publications takes attention away from the need to focus on all types of unethical contributions in peer-reviewed publications, irrespective of the source of the unethical practice.

The various definitions of ghostwriting and ghost authoring found in this systematic review highlights the considerable disagreement in the medical literature with regard to the definitions of ghostwriting and ghost authoring and whether a distinction should be made between these unethical practices. Although the Council of Science Editors uses a straightforward and broad definition of ghost authors that does not distinguish between ghost authors and ghostwriters,[2] the distinction between ghost authorship and ghostwriting used in this manuscript is consistent with those used in the cross-sectional surveys conducted by Flanagin[67] and Wislar[68] and with the authorship criteria recommended by the ICMJE.[12] Distinguishing between these practices is important because it differentiates between undisclosed contributions that do and do not merit authorship. However, part of the reason

for the disagreement about definitions of ghost authoring and ghostwriting stems from differences of opinion in the medical literature over how authorship is defined, particularly with regard to what constitutes “substantial” contributions to a publication and the role of disclosure of contributions to a manuscript in the acknowledgments.[69] The ICMJE criteria for authorship[12] are the most well recognised authorship criteria available, but leave much about authorship undefined.[63, 70] Despite this, the recent emphasis on author accountability in the ICMJE’s fourth criterion is an attempt to better define authors as those persons who are responsible and accountable for the content of a published work. While a professional medical writer can take responsibility for the writing and how research is reported in a manuscript, the professional medical writer cannot take responsibility for the integrity of the research or be accountable for the clinical interpretation of the findings, unless he or she was involved in the generation of the research or its analysis. Responsibility for the integrity of research and accountability for the clinical interpretation of the findings is, and should always be, the responsibility of the authors. Until a more definitive model of authorship is universally accepted, the controversy and disagreement over definitions of authors, ghostwriters, and ghost authors will remain. However, no matter what term is used, the practice of misleading readers about potential competing interests and hiding contributions to a published work, no matter whether these contributions should be most appropriately disclosed in the author byline or acknowledgements section, is unethical and totally unacceptable.

The strengths of this systematic review are that a broad search strategy was employed with few restrictions to minimize any potential for publication or language bias. All study designs and publication types were considered and there were no restrictions on language. Although the full text or abstracts of 8 publications could not be retrieved, omission of these publications was unlikely to have biased the findings. Review of the abstracts or titles of these publications suggested that none reported numerical estimates of the prevalence of ghostwriting. The major finding of this review was the limitations of the reported evidence of

ghostwriting in the medical literature. These limitations included the heterogeneity among the studies in the outcomes reported and populations investigated, the observational study designs, and the retrospective nature of data collection. Given the nature of unethical authorship practices, it may not be feasible to conduct a prospective study on ghostwriting. However, based on the findings from this review, recommendations can be made to help researchers, authors, editors, and peer-reviewers apply the same rigorous standards that are applied to the conduct and assessment of all clinical research and actively improve the quality of reporting of the evidence of unethical authorship practices.

Researchers should use a standard definition of ghostwriting so that the confusion around ghostwriters and ghost authors is not perpetuated. For example, the descriptive analysis of Danish industry-initiated trials used a nonstandard definition of authorship, which is likely to have contributed to the very high prevalence of possible ghostwriting reported in this study (75%). In this study, Gøtzsche et al.[30] suggested that individuals who write the trial protocol, conduct the statistical analyses, or who contribute to the writing of a publication should be included as authors. Indeed, the prevalence of ghost authorship in this study was 91% when the analyses included these individuals, irrespective of whether they had been appropriately acknowledged elsewhere. Given that the Gøtzsche et al.[30] study has been downloaded more than 3,000 times since publication[71] and cited repeatedly as evidence not only of ghost authorship but also of ghostwriting,[40, 44, 46, 48] it would have been illuminating if the authors had included an estimate of the prevalence of ghostwriting using standard definitions for comparison. In addition to using standard definitions of ghostwriting, researchers and authors should ensure that cited evidence of ghostwriting is reported accurately without unwarranted generalizations. Publications based on personal commentary should be avoided and studies that use nonstandard definitions, specific populations, or that were conducted before a change in practice (eg, before the adoption of the Good Publication Practice guidelines for communicating company sponsored research in 2003 [GPP][72] and in 2009 [GPP2][11]) should be described in context.

Editors and peer reviewers have a duty to prevent outdated, misleading, or mistaken evidence on the prevalence of ghostwriting from being published and perpetuated. Close attention should be paid to the internal and external validity of the study, the definitions used, how the data are reported, and whether the data are interpreted within the context of current practices. When assessing a submitted manuscript on ghostwriting, editors should consider using peer reviewers with expertise in the appropriate study designs (eg, survey research) and ethical publication practices. Collectively, these actions could help prevent further questionable evidence on the prevalence of ghostwriting from being published.

In conclusion, the findings from this systematic review showed that reports of the prevalence of ghostwriting in the medical literature are limited by the varied definitions used to describe unethical authorship practices, the types of study designs employed, and the populations assessed. To improve reporting, researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. While open and transparent debate should be encouraged, editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

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CONTRIBUTORSHIP STATEMENT

The author, SS, contributed to the conception and design of the systematic literature review, the collection and screening of publications, and the analysis and interpretation of the findings. SS drafted and critically revised draft versions of the manuscript and gave final approval for submission.

COMPETING INTERESTS

The author is a professional medical writer who is actively involved in national and international not-for-profit organizations that encourage ethical medical writing practices. The author has no relationships with any pharmaceutical company that might have an interest in the submitted work and has no other relationships that could be construed as a competing interest.

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DATA SHARING

All data associated with the study are fully available to the author.

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FIGURE LEGENDS

Figure 1. Selection of publications reporting a numerical estimate of the prevalence of ghostwriting. Electronic databases were searched on 23 May 2013.

[a] Journal of the European Medical Writers Association, formerly known as The Write Stuff.

[b] Journal of the American Medical Writers Association.

[c] Council of Science Editors Annual Meetings.

[d] International Congress on Peer Review and Biomedical Publication.

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[6] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.

TITLE

Systematic review on the primary and secondary reporting of the prevalence of ghostwriting in the medical literature

AUTHOR

Serina Stretton, PhD CMPP

AUTHOR'S AFFILIATION

ProScribe – Envision Pharma Group ~~Medical Communications~~, ~~Noosaville, Queensland~~Sydney, NSW, Australia

AUTHOR FOR CORRESPONDENCE

Serina Stretton, PhD, CMPP

ProScribe – Envision Pharma Group~~Medical Communications~~

18 Poulet St

Matraville, NSW, 2036

AUSTRALIA

Tel: + 612 9661 0768

Fax: + 61~~2~~ 7 5474 9156~~9661 0766~~

Email: serina.stretton@EnvisionPharmaGroup.com~~ss@proscribe.com.au~~

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Authorship, ghostwriting, systematic review

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ABSTRACT

Background: Ghostwriting of industry-sponsored articles is unethical and is perceived to be common practice.

Objective: To systematically review how evidence for the prevalence of ghostwriting is reported in the medical literature.

Data sources: MEDLINE via PubMed 1966+, EMBASE 1966+, The Cochrane Library 1988+, Medical Writing 1998+, The AMWA Journal 1986+, Council of Science Editors Annual Meetings 2007+, and the Peer Review Congress 1994+ were searched electronically (23 May 2013) using the search terms ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*.

Eligibility criteria: All publication types were considered; only publications reporting a numerical estimate of possible ghostwriting prevalence were included.

Data extraction: Two independent reviewers screened the publications; discrepancies were resolved by consensus. Data to be collected included a numerical estimate of the prevalence of possible ghostwriting (primary outcome measure), definitions of ghostwriting reported, source of the reported prevalence, publication type and year, study design, and sample population.

Results: Of the 848 publications retrieved and screened for eligibility, 48 reported numerical estimates for the prevalence of possible ghostwriting. Sixteen primary publications reported findings from cross-sectional surveys or descriptive analyses of published articles; 32 secondary publications cited published or unpublished evidence. Estimates on the prevalence of possible ghostwriting in primary and secondary publications varied markedly. Primary estimates were not suitable for meta-analysis because of the various definitions of ghostwriting used, study designs, and types of populations or samples. Secondary estimates were not always reported or cited correctly or appropriately.

Conclusions: Evidence for the prevalence of ghostwriting in the medical literature is limited and can be outdated, misleading, or mistaken. Researchers should not inflate estimates

using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. Editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

Strengths

- First systematic review on the reporting of the prevalence of ghostwriting in the medical literature
- A broad search strategy was employed with few restrictions to minimize any potential for publication or language bias; there were no restrictions on language.
- All study designs and publication types were considered

Limitations

- Study heterogeneity in the outcomes reported and populations investigated precluded synthesis of the data
- Retrospective and self-reported nature of the data collection increased the risk of selection bias in the studies
- Most included studies were not broadly representative of the peer-reviewed medical literature

INTRODUCTION

Ghostwriting occurs when paid or unpaid writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments.[1] This practice is considered to be distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[1] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding ~~the two~~these unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[2]

Ghostwriting of peer-reviewed journal publications in the medical literature is believed to be common practice.[3-5] This belief is supported, to a large extent, by highly publicized cases, primarily from the 1990s and early 2000s, of pharmaceutical companies and authors who had used ghostwriters to prepare manuscripts for publication in medical journals.[6-8] Such cases are highly unethical because the role of the commercial sponsor and any other potential conflicts of interest were hidden.

~~Ghostwriting occurs when writing contributions to a manuscript that do not meet authorship criteria are not disclosed in the acknowledgments. This practice is distinct from ghost authoring, where contributions to a manuscript that do merit authorship are not disclosed in the author byline.[7] However, part of the challenge in understanding the prevalence of ghostwriting is the current confusion surrounding the two unethical practices. For example, the widely used Council of Science Editors definition of ghost authors (ie, individuals who participate in research, data analysis, and/or writing of a manuscript but are not named or disclosed in the author byline or acknowledgments) does not distinguish between ghost authors and ghostwriters.[8]~~ Estimating the prevalence of ghostwriting in the medical

literature has ~~also~~ been hindered not only by the different definitions of ghostwriting and ghost authoring, but also by the failure of authors to distinguish ghostwriting, which is unethical, from professional medical writing support, which is ethical.[9, 10] Ghostwriters keep their involvement in a manuscript hidden, whereas professional medical writers disclose their involvement and follow ethical publication practices.[1, 9-11] Consistent with the authorship criteria recommended by the International Committee of Medical Journal Editors (ICMJE), professional medical writers who provide writing assistance and do not meet all of the ICMJE authorship criteria, should be acknowledged rather than listed as authors.[12] The prevalence of disclosed professional medical writing assistance in medical journals has been estimated to be between 6.0% and 11.0%[10, 13] and the legitimate value that this medical writing assistance can bring to improving the quality, timeliness, and integrity of reporting in medical journals has been demonstrated.[1, 14, 15] However, the exact prevalence of ghostwriting and other forms of undisclosed contributions to papers published in medical journals is unknown.

The primary objective of this study was to conduct the first systematic review on how the prevalence of ghostwriting is reported in the medical literature. The secondary objectives were to assess the variability of the reported estimates of the prevalence of ghostwriting and investigate the source for these estimates.

MATERIALS AND METHODS

Literature search strategy

A search strategy was developed to retrieve publications from the medical literature that reported quantitative estimates of the prevalence of ghostwriting. The following databases were searched on 23 May 2013: MEDLINE via PubMed (1966+); EMBASE (1966+); The Cochrane Library, including the Cochrane Database of Systematic Reviews and Cochrane Central Register of Controlled Trials (1988+); The AMWA Journal (1986+, journal of the American Medical Writers Association); Medical Writing (1998+, journal of the European Medical Writers Association, formerly known as The Write Stuff); Council of Science Editors annual meetings (2007+); and the International Congress on Peer Review and Biomedical Publication meetings (1994+). General text was searched using the following search terms: ghostwrit*, ghostauthor*, ghost AND writ*, ghost AND author*. Truncation symbols and Boolean operators (AND, OR) were used wherever possible.

Two reviewers (SS, SMG) independently screened the title and abstracts of all retrieved publications using prespecified eligibility criteria; the full text of potentially eligible publications was screened to confirm eligibility for inclusion. Any discrepancies between the reviewers were resolved by consensus. The reference lists of relevant reviews and other publications were screened by hand to identify any additional publications for inclusion.

Eligibility criteria

The eligibility criteria were prespecified. Publications were included if they reported a numerical estimate of the prevalence of possible ghostwriting. Publications were excluded if they were duplicate publications from different databases or abstracts of subsequently published full-text articles, did not report any outcomes related to ghostwriting, or reported qualitative estimate(s) of the prevalence of possible ghostwriting. No restrictions on language were included in the eligibility criteria. If needed, the abstracts of publications

written in non-English language were to be translated to assist in screening for eligibility. If publications were eligible for inclusion, the full text of the publications were to be translated to English.

Data extraction and analysis

The primary and secondary outcomes and dData to be collected were prespecified. Data to be collected and included publication type and year, study design and sample population, definitions of ghostwriting reported, reported prevalence of possible ghostwriting, and the source of the reported prevalence of ghostwriting. The prevalence of possible ghostwriting was reported as published. No unpublished data from the retrieved literature were reported and values for prevalence were not extrapolated from published data.

The primary outcome measure for the systematic review was a numerical estimate of the prevalence of possible ghostwriting. The reported definitions of possible ghostwriting were categorized according to the following standard definitions, irrespective of the term used to describe the practice in the publication. Undisclosed writing contributions to a manuscript were defined as (i) ghostwriting if they were described as not meriting authorship and were not listed in the acknowledgments[9, 16] and (ii) ghost authoring if they did merit authorship and were not listed in the author byline.[1] Consistent with recommendations from international medical journal editors,[12, 14] Good Publication Practice guidelines,[11] and professional medical writing associations,[17, 18] disclosed writing contributions to a manuscript that did not merit authorship and were disclosed in the acknowledgments were not categorized as ghostwriting.

The key factors considered when assessing study quality were study design, the population assessed, and how ghostwriting was defined. While there is no consensus on the best practice for reporting survey research,[19] the quality of the cross-sectional surveys was

determined by an assessment of the following factors: validation or pretesting of the survey questions, anonymity of the response, sample size, and response rate.

RESULTS

Publication selection

A total of 848 articles were retrieved from the literature search, 800 were excluded, and 48 met the eligibility criteria and were included in the systematic review (Figure 1). The main reasons for exclusion were publications not relevant to ghostwriting (n = 539), duplicate publications from different databases (n = 129), and publications not reporting a numerical estimate of the prevalence of possible ghostwriting (n = 124). The titles or abstracts of the 8 publications that were excluded because the full text or abstract could not be retrieved were reviewed; none were considered to report a numerical estimate of the prevalence of possible ghostwriting. Overall, eligible publications included 16 primary publications that reported original research on the prevalence of possible ghostwriting and 32 secondary publications that cited published or unpublished evidence of possible ghostwriting.

Primary publications

Publication characteristics

Of the 16 primary publications (Table 1), there were 13 full-text publications[6, 8, 20-30] and three conference abstracts[31-33] that reported a numerical estimate of possible ghostwriting. Twelve publications[20-28, 31-33] reported findings from cross-sectional surveys and four publications[6, 8, 29, 30] reported findings from descriptive analyses of published articles. ~~While there is no consensus on the best practice for reporting survey research,[33]~~M~~ost~~ost cross-sectional surveys were reasonably well reported. The surveys used were not validated but most included pretested questions, required an anonymous response, were conducted in targeted populations (ie, all individuals invited to participate were involved in the preparation of peer-reviewed manuscripts), and reported a sample size and response rate. Of the cross-sectional surveys conducted in targeted populations (eg, corresponding or first authors), six had response rates greater than 50%, two had response rates less than 50%, and one did not report a

response rate (Table 1). Of the cross-sectional surveys conducted in nontargeted populations (eg, individuals invited to participate who may or may not have been involved in providing medical writing assistance for peer-reviewed publications), three had low (12% to 28%) response rates and one did not report a response rate (Table 1).

Table 1. Characteristics of Primary Publications Reporting Original Evidence of Ghostwriting

Publication	Population	Survey description	Data collected	N	RR
Cross-sectional surveys – authors or corresponding authors					
Flanagin 1998[27]	6 general medicine journals [a]	Self-administered, postal, anonymous, pretested, targeted	1996	809	69%
Mowatt 2002[23]	Published Cochrane reviews	Self-administered, online, anonymous, pretested, targeted	1999	362	63%
Hao 2009[32] (Abstract)	Chin Med J	Self-administered, email, not anonymous, targeted	2008 [c]	220	86%
Dotson 2011[28]	3 pharmacy journals	Self-administered, online, anonymous, pretested, targeted	2009	112	25%
Mirzazadeh 2011[24]	3 Iranian journals	Self-administered, email, not anonymous, targeted	2009-2010	NR	NR
Wislar 2011[20]	6 general medicine journals [b]	Self-administered, online, anonymous, pretested, targeted	2008	622	70%
Ghajarzadeh 2012[26]	Arch Iran Med (based on student theses)	Self-administered, email, anonymous, targeted	2005-2007	30	49%
Vinther 2012[21]	Ugeskr Laeger & Dan Med J	Self-administered, online, anonymous, pretested, targeted	2010	272	62%
Cross-sectional surveys – healthcare professionals					
Price 2000[22]	Health academic staff	Self-administered, postal, anonymous, pretested, targeted	NR	166	59%
Rees 2013[31]	Registered users of	Self-administered, online,	NR	295	NR

(Abstract)	EPG Online	anonymous, nontargeted			
Cross-sectional surveys – members of medical writing associations					
Jacobs 2009[25]	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2005 2008	843 773	28% 14%
Hamilton 2012[33] (Abstract)	EMWA / AMWA members	Self-administered, online, anonymous, pretested, nontargeted	2011	620	12% [d]
Publication reviews and descriptive analysis					
Healy 2003[6]	Articles on sertraline	NA	1998-2000	96	NA
Gøtzsche 2007[30]	Articles on Danish industry-initiated trials approved 1994-1995	NA	NR	44	NA
Ross 2008[8]	Reviews on rofecoxib associated with Merck support	NA	1996-2004	72	NA
Suda 2011[29]	Noninferiority clinical trials	NA	1989-2009	583	NA

[a] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, American Journal of Cardiology, American Journal of Medicine, American Journal of Obstetrics and Gynecology.

[b] Annals of Internal Medicine, JAMA, the New England Journal of Medicine, Lancet, Nature Medicine, PLoS Medicine.

[c] Submission date.

[d] Personal communication, C. Hamilton.

Abbreviations: AMWA, American Medical Writers Association; EMWA, European Medical Writers Association; NA, not applicable; NR, not reported; RR, response rate.

Not all cross-sectional surveys were broadly representative of the peer-reviewed medical literature. The cross-sectional surveys were conducted in single populations of academic staff or healthcare professionals[22, 31], members of medical writing associations[25, 33], or corresponding or first authors. The corresponding or first authors were surveyed from single journals (Cochrane reviews, Chinese Medical Journal, Archives of Iranian Medicine),[23, 26, 32] two Danish journals,[21] three Iranian journals,[24] and three pharmacy journals.[28] Two cross-sectional surveys of corresponding authors were conducted in six general medicine journals.[20, 27]

The descriptive analyses of published articles were conducted in single populations and included an analysis of sertraline publications from 1998 to 2000,[6] rofecoxib reviews from 1996 to 2004,[8] publications from industry-initiated trials in Denmark from 1994 to 1995,[30] and from published noninferiority trials from 1989 to 2009.[29]

Reported prevalence of ghostwriting

The reported prevalence of possible ghostwriting in the primary publications varied markedly and were not suitable for meta-analysis~~and was difficult to compare~~ because of the different populations assessed, the different methods used to generate the estimates, and the various definitions that were used (Tables 1 and 2). All cross-sectional surveys reported a definition of ghostwriting or ghost authoring, but most definitions did not differentiate contributions that merited authorship from those that did not merit authorship (Table 2).

Table 2. Primary Publications Reporting Estimates of the Prevalence of Possible Ghostwriting

Publication	Measure of Possible Ghostwriting Reported ~~measure~~by Authors	Estimate % (n)
Cross-sectional surveys reporting ghostwriting		
Flanagin 1998[27]	Unnamed individual who participated in the writing	1.4% (11/809) of articles
Wislar 2011[20]	Unnamed individual who participated in the writing	0.2% [a] (1/622) of articles
Jacobs 2009, Hamilton 2012[25, 33]	Undisclosed medical writing assistance not qualifying for authorship	2005: 61.8% (NR) 2009: 41.7% (NR) 2011: 33.0% (NR) of ~~writers~~articles [b]
Cross-sectional surveys reporting combined ghost authoring and ghostwriting		
Flanagin 1998[27]	Failure to name, as an author, individuals who made substantial contributions to the research or writing or an unidentified medical writer	11.5% (93/809) of articles
Price 2000[22]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	24.1% (40/166) of authors
Mowatt 2002[23]	Individual merited authorship or had assisted with drafting but not listed as an author or acknowledged	8.8% (32/362) of articles
Hao 2009[32]	English-language speakers assisted with writing but not identified as authors or acknowledged	10.4% (NR) of authors
Dotson 2011[28]	Failure to name, as an author, individuals who made substantial contributions to the research or writing	0.9% (1/112) of articles
Wislar 2011[20]	Failure to name, as an author, individuals who made substantial contributions to the research or writing of the article or an unnamed individual who participated in the writing	7.9% (49/622) of articles
Cross-sectional surveys reporting ghost authoring		
Mirzazadeh 2011[24]	Failure to name, as an author, individuals who made substantial contributions to the research	21.4% (25/NR) of authors

Ghajarzadeh 2012[26]	Failure to name, as an author, students who made substantial contributions to the research [c]	0.7% (2/296) of articles
Vinther 2012[21]	Individual merited authorship but not listed as an author	2.4% (6/245) of articles
Rees 2013[31]	Individual merited authorship but not listed as an author	70% (NR/202) of published authors
Publication reviews and descriptive analysis reporting possible ghost authoring or ghostwriting		
Healy 2003[6]	Published articles coordinated by a medical information company, including acknowledged medical writing support [d~~c~~]	57.3% (55/96) of articles
Gøtzsche 2007[30]	Individuals who wrote the trial protocol, conducted the statistical analyses, or wrote the manuscript but were not listed as authors, not members of a study group or steering committee, or not disclosed in an acknowledgment	75.0% (33/44) of trials
Ross 2008[8]	Published reviews associated with Merck support and with a single external author [e~~d~~]	69.4% (50/72) of reviews

[a] Available as online supplementary data.

[b] Values represent the mean weighted percentage of publications that were ghostwritten by respondents. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

[b~~,~~ c] Students were classified as ghostwriters if the student was not named as an author and if the results reported in the publications were based on the results of their theses.

[c~~,~~ d] Authors conclude data provide quantification of the possible extent of ghostwriting based on a single drug. Of the 55 published articles that were coordinated through a medical information company, 2 included medical writing assistance that was acknowledged in the published article.

[d~~,~~ e] Published review articles had been identified from correspondence between Merck and a medical publishing company, from Merck publication status reports, or were affiliated with an author named within the correspondence or publication status reports. The authors did not report whether medical writing assistance was acknowledged in the published article.

Abbreviations: NR, not reported.

Four cross-sectional surveys reported the prevalence of ghostwriting where the definition provided could be categorized as undisclosed contributions that do not merit authorship.[20, 25, 27, 33] Findings from these surveys, which were repeated at different time points, suggested that the prevalence of ghostwriting is low and decreasing. Flanagin[27] and Wislar[20] conducted two similar cross-sectional surveys of corresponding authors of articles published in six general medicine journals in 1996[27] and in 2008.[20] The surveys, which included a core set of three journals (Annals of Internal Medicine, JAMA, the New England Journal of Medicine) at each time point and in targeted populations, used pretested questions that required an anonymous response and had response rates greater than 65%. The prevalence of ghost authoring (which included ghostwriting) was estimated to be 11.5% in 1996 and 7.9% in 2008. The prevalence of ghostwriting from these surveys was estimated to be 1.4% in 1996 and 0.2% in 2008. Hamilton and Jacobs conducted a cross-sectional survey of members of two medical writing associations in 2005, 2008, and 2011.[25, 33] The survey, which was not conducted in targeted populations, used pretested questions that required an anonymous response and had response rates from 12% to 28% of the total population surveyed (ie, not all participants surveyed were involved in the preparation of peer-reviewed manuscripts). Findings from these surveys showed that the mean weighted percentage of publications that were ~~association members who contributed to peer-reviewed publications and had~~ ghostwritten by respondents ~~at least once during the survey year~~ had decreased from 61.8% in 2005 to 33.0% in 2011. Findings were weighted in proportion to the number of manuscripts the respondent wrote per year.

Six cross-sectional survey publications reported the prevalence of possible ghostwriting (ie, the prevalence of ghostwriting and ghost authoring combined).[20, 22, 23, 27, 28, 32] The prevalence of possible ghostwriting reported in these cross-sectional surveys varied from 0.9% to 24.1% of publications or authors.

Four cross-sectional survey publications reported the prevalence of ghost authoring.[21, 24, 26, 31] The prevalence of ghost authoring reported in these cross-sectional surveys varied from 0.7% to 70% of publications or authors.

The descriptive analyses of published articles on sertraline and rofecoxib did not include a prespecified definition of ghost authoring or ghostwriting;[6, 8] in these studies, possible ghostwriting or ghost authoring was assumed in publications associated with industry-sponsored support (Table 2). The descriptive analysis of industry-initiated trials in Denmark used a nonstandard definition of ghost authors; in this study ghost authors were defined as individuals, not named as authors, who were involved in writing the protocol, conducting the statistical analyses, or writing the manuscript.[30] The prevalence of possible ghostwriting in the two descriptive analyses of single drugs that did not include a prespecified definition of ghostwriting was 57% of articles on sertraline published from 1998 to 2000[6] and 69% of reviews on rofecoxib published from 1996 to 2004.[8] The prevalence of possible ghostwriting in the descriptive analysis of Danish industry-initiated trials, which used a nonstandard definition of authorship, was 75% of Danish initiated trials approved in 1994 to 1995.[30] The descriptive analysis of noninferiority trials[29] was not considered further as the definition of ghostwriting used was consistent with disclosed medical writing assistance. In this study ghostwriters were defined as acknowledged individuals, other than authors, who contributed to the writing and were affiliated with an industry-sponsored study.[29] The prevalence of disclosed medical writing assistance in this descriptive analysis was 17.3% (101/583) of clinical trials.

Secondary publications

Publication characteristics

Of the 32 secondary publications, there were 13 review articles, 10 editorials, 5 commentaries, 3 news articles, and 1 government report, with most being published after 2008 (Table 3). Most publications cited primary sources as evidence of the prevalence of

possible ghostwriting (Table 3), with the cited prevalence of possible ghostwriting varying from 6% of publications to 100% of publications involving drugs. In most secondary publications, the information on the prevalence of ghostwriting was not reported consistently compared with the cited evidence (Table 3). Misleading and mistaken information was reported in many publications that (i) did not distinguish between ghostwriting and ghost authoring, (ii) included acknowledged medical writing assistance or a combined estimate of guest authorship and ghost authorship as ghostwriting, (iii) generalized findings from publication reviews and analyses of specific datasets to wider populations of publications or industry-sponsored trials, or (iv) cited personal communications or informal surveys where the original source was unpublished and could not be verified (Table 3).

Table 3. Characteristics of Secondary Publications Citing Evidence of Possible Ghostwriting

Publication	Cited evidence	Consistent with cited source(s)	Comment
Elliot Hastings Cent Rep 2004[34]	Primary	No	Did not distinguish GW from GA
Moffat Perspect Biol Med 2007[35]	Primary	No	Did not distinguish GW from GA
Schiefe Pharmacotherapy 2009[36]	Primary	No	Did not distinguish GW from GA
MacLennan Climacteric 2010[37]	Primary	No	Did not distinguish GW from GA
Nahai Aesthet Surg J 2010[38]	Secondary	No	Did not distinguish GW from GA
Ngai Account Res 2005[39]	Primary	Yes	Did not distinguish GW from GA
Bosch EMBO Rep 2011[40]	Primary	Yes	Did not distinguish GW from GA
Wiwanitkit Am J Med 2012[41]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Krimsky Med Law 2007[42]	Primary	No	Did not distinguish GW from GA or GA from guest authorship
Langdon-Neuner Mens Sana Monogr 2008[43]	Primary & secondary	No	Did not distinguish GW from GA or GW from disclosed medical writing support

Tharyan Indian J Med Ethics 2011[44]	Primary	Yes	Did not distinguish GW from GA or GW from disclosed medical writing assistance
Paul Clin Microbiol Infect 2009[45]	Primary	Yes	Did not distinguish GW / GA from disclosed medical writing assistance
Bavdekar Lung India 2012[46]	Primary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Górski Transplant Proc 2010[47]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
Matias-Guiu Neurologia 2011[48]	Primary & secondary	No	Did not distinguish GW from GA and generalized evidence to a wider population
McHenry Mens Sana Monogr 2010[49]	Gov. report	No	Generalized evidence to a wider population
Healy BMJ 2004[50]	Primary & secondary	No / ND	Generalized evidence to a wider population and reported personal opinion of GW prevalence
Abbasi BMJ 2004[4]	Secondary	No	Secondary publication cited named individual
Mitrany Science Editor 2005[51]	None	ND	Cited named individual
Collier Can Med Assoc J 2009[5]	None	ND	Cited named individual
Kmietowicz BMJ 2004[52]	None	ND	Cited named individual
Matthews Wall St J 2005[3]	None	ND	Cited unpublished data
Bonita Heart Fail Clin 2011[53]	Primary	Yes	Consistent with cited source
Grassley Int J Occup Environ Health 2011[54]	Primary	Yes	Consistent with cited source

Nat Clin Pract Nephrol 2007[55]	Primary	Yes	Consistent with cited source
Editors J Urol 2008[15]	Primary	Yes	Consistent with cited source
Baethge Deutsches Arzteblatt 2009[56]	Primary	Yes	Consistent with cited source
Flanagin CSE Annual Mtg 2010[57]	Primary	Yes	Consistent with cited source
Murray Open Med 2010[58]	Primary	Yes	Consistent with cited source
Moore BMJ 2004[59]	Secondary	Yes	Consistent with cited source
Hargreaves BMJ 2007[60]	Primary	Yes	Consistent with cited source
Jones Nature 2009[61]	Primary	Yes	Consistent with cited source

Abbreviations: CSE, Council of Science Editors; GA, ghost authoring; Gov, government; GW, ghostwriting; ND, not determined.

Case study of misleading and mistaken evidence

An analysis of secondary publications citing the possible prevalence of ghostwriting in sertraline publications provides an illuminating case study on how misleading or mistaken evidence can enter and remain in the medical literature. In 2003, Healy and Cattell showed that 57% (55/96) of articles on sertraline published from 1998 to 2000 were coordinated through a medical information company and only two acknowledged medical writing assistance.[6] The authors concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". In 2004 (October 14), Healy also provided evidence on the influence of the pharmaceutical industry on key groups at a United Kingdom House of Commons Health Select Committee investigation.[62] In answer to Question 197, Healy stated the following:

"My estimate is that, even in journals like the BMJ, the Lancet, the New England Journal of Medicine and JAMA, the leading journals in the field, if these articles have to do with therapeutics, with drugs, it may be worse perhaps for psychiatry than elsewhere, but I doubt it, 50% of these articles are ghostwritten. It may be higher."

Healy and Cattell's original evidence has been cited incorrectly and interchangeably with Healy's statement to the House of Commons Health Select Committee investigation (Figure 2). Although findings from the primary publication have been cited and interpreted correctly in two secondary publications,[34, 35] many secondary publications incorrectly cited Healy's original evidence and statement to the House of Commons Health Select Committee investigation (Figure 2).

DISCUSSION

This systematic review on the reported prevalence of ghostwriting in the medical literature showed that the estimates of the prevalence of ghostwriting in primary publications varied markedly and were influenced by the definitions used, the types of study designs, and the type of population or samples assessed. In addition, secondary publications often cited outdated, misleading, or mistaken evidence of the reported prevalence of ghostwriting, with many publications not distinguishing ghostwriting from ghost authoring.

Although evidence from descriptive analyses can highlight the extent of ghostwriting in single populations, evidence from well-conducted cross-sectional surveys have the potential to provide estimates of the prevalence of ghostwriting that may be generalized to the majority of peer-reviewed publications. Despite this, many of the cross-sectional surveys retrieved in this systematic review were conducted in limited populations that were not broadly representative of the peer-reviewed medical literature. In addition, many of the cross-sectional surveys did not differentiate between contributions that merited authorship from those that did not merit authorship and provided at best, an estimate of possible ghostwriting. The reported prevalence of ghostwriting, where ghostwriting was defined as undisclosed contributions that did not merit authorship, was retrieved from the two cross-sectional surveys of corresponding authors from several general medicine journals and by cross-sectional surveys (repeated on three separate occasions from 2005 to 2011) of members of two major medical writing associations. Together the findings from these surveys suggested that the prevalence of ghostwriting has decreased in recent years. However, while the findings from these surveys may be considered more broadly representative of the peer-reviewed medical literature than surveys focused on single journals, single countries, or single subject areas, interpretation of these findings should take into account that respondents were required to retrospectively self-report potentially unethical or unprofessional behaviour.

Unethical authorship practices are a major concern and are an increasingly recognized problem in the medical literature.[63] As the findings from this systematic review suggest, some of these perceived problems may arise from the considerable, but unnecessary, confusion and disagreement surrounding the definitions of ghostwriting. As recognized by the World Association of Medical Editors, professional medical writers can have a legitimate role in assisting authors to communicate their research findings in the peer-reviewed literature.[14] Professional medical writers can and do improve the timeliness and quality of reporting and can assist investigators and industry sponsors to meet their ethical commitments to the disclosure and publishing of clinical trial results.[1, 64-66] As such, the misleading and mistaken reporting of the prevalence of ghostwriting that was evident in the secondary publications retrieved in this systematic review is disappointing. Specifically, the confusion surrounding definitions of ghost authorship and ghostwriting and the unbalanced focus on industry as the source of unethical authorship practices in the secondary publications takes attention away from the need to focus on all types of unethical contributions in peer-reviewed publications, irrespective of the source of the unethical practice.

The various definitions of ghostwriting and ghost authoring found in this systematic review highlights the considerable disagreement in the medical literature with regard to the definitions of ghostwriting and ghost authoring and whether a distinction should be made between these unethical practices. Although the Council of Science Editors uses a straightforward and broad definition of ghost authors that does not distinguish between ghost authors and ghostwriters,[2] the distinction between ghost authorship and ghostwriting used in this manuscript is consistent with those used in the cross-sectional surveys conducted by Flanagin[67] and Wislar[68] and with the authorship criteria recommended by the ICMJE.[12] Distinguishing between these practices is important because it differentiates between undisclosed contributions that do and do not merit authorship. However, part of the reason

for the disagreement about definitions of ghost authoring and ghostwriting stems from differences of opinion in the medical literature over how authorship is defined, particularly with regard to what constitutes "substantial" contributions to a publication and the role of disclosure of contributions to a manuscript in the acknowledgments.[69] The ICMJE criteria for authorship[12] are the most well recognised authorship criteria available, but leave much about authorship undefined.[63, 70] Despite this, the recent emphasis on author accountability in the ICMJE's fourth criterion is an attempt to better define authors as those persons who are responsible and accountable for the content of a published work. While a professional medical writer can take responsibility for the writing and how research is reported in a manuscript, the professional medical writer cannot take responsibility for the integrity of the research or be accountable for the clinical interpretation of the findings, unless he or she was involved in the generation of the research or its analysis. Responsibility for the integrity of research and accountability for the clinical interpretation of the findings is, and should always be, the responsibility of the authors. Until a more definitive model of authorship is universally accepted, the controversy and disagreement over definitions of authors, ghostwriters, and ghost authors will remain. However, no matter what term is used, the practice of misleading readers about potential competing interests and hiding contributions to a published work, no matter whether these contributions should be most appropriately disclosed in the author byline or acknowledgements section, is unethical and totally unacceptable.

The strengths of this systematic review are that a broad search strategy was employed with few restrictions to minimize any potential for publication or language bias. All study designs and publication types were considered and there were no restrictions on language. Although the full text or abstracts of 8 publications could not be retrieved, omission of these publications was unlikely to have biased the findings. Review of the abstracts or titles of these publications suggested that none reported numerical estimates of the prevalence of ghostwriting. The major finding of this review was the limitations of the reported evidence of

ghostwriting in the medical literature. These limitations included the heterogeneity among the studies in the outcomes reported and populations investigated, the observational study designs, and the retrospective nature of data collection. Given the nature of unethical authorship practices, it may not be feasible to conduct a prospective study on ghostwriting. However, based on the findings from this review, recommendations can be made to help researchers, authors, editors, and peer-reviewers apply the same rigorous standards that are applied to the conduct and assessment of all clinical research and actively improve the quality of reporting of the evidence of unethical authorship practices.

Researchers should use a standard definition of ghostwriting so that the confusion around ghostwriters and ghost authors is not perpetuated. ~~Ghostwriting is paid or unpaid writing assistance by individuals who generally do not merit authorship and whose contributions are not disclosed in the acknowledgments.[1] In contrast, ghost authoring is contributions to the research, data analysis, and/or writing of a manuscript by individuals who do merit authorship and whose contributions are not disclosed in the author byline.[1]~~ For example, the descriptive analysis of Danish industry-initiated trials used a nonstandard definition of authorship, which is likely to have contributed to the very high prevalence of possible ghostwriting reported in this study (75%). In this study, Gøtzsche et al.[30] suggested that individuals who write the trial protocol, conduct the statistical analyses, or who contribute to the writing of a publication should be included as authors. Indeed, the prevalence of ghost authorship in this study was 91% when the analyses included these individuals, irrespective of whether they had been appropriately acknowledged elsewhere. Given that the Gøtzsche et al.[30] study has been downloaded more than 3,000 times since publication[71] and cited repeatedly as evidence not only of ghost authorship but also of ghostwriting,[40, 44, 46, 48] it would have been illuminating if the authors had included an estimate of the prevalence of ghostwriting using standard definitions for comparison. In addition to using standard definitions of ghostwriting, researchers and authors should ensure that cited evidence of ghostwriting is reported accurately without unwarranted generalizations. Publications based

on personal commentary should be avoided and studies that use nonstandard definitions, specific populations, or that were conducted before a change in practice (eg, before the adoption of the Good Publication Practice guidelines for communicating company sponsored research in 2003 [GPP][72] and in 2009 [GPP2][11]) should be described in context.

Editors and peer reviewers have a duty to prevent outdated, misleading, or mistaken evidence on the prevalence of ghostwriting from being published and perpetuated. Close attention should be paid to the internal and external validity of the study, the definitions used, how the data are reported, and whether the data are interpreted within the context of current practices. When assessing a submitted manuscript on ghostwriting, editors should consider using peer reviewers with expertise in the appropriate study designs (eg, survey research) and ethical publication practices. Collectively, these actions could help prevent further questionable evidence on the prevalence of ghostwriting from being published.

In conclusion, the findings from this systematic review showed that reports of the prevalence of ghostwriting in the medical literature are limited by the varied definitions used to describe unethical authorship practices, the types of study designs employed, and the populations assessed. To improve reporting, researchers should not inflate estimates using nonstandard definitions of ghostwriting nor conflate ghostwriting with other unethical authorship practices. While open and transparent debate should be encouraged, editors and peer reviewers should not accept articles that incorrectly cite or interpret primary publications that report the prevalence of ghostwriting.

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COMPETING INTERESTS

The author is a professional medical writer who is actively involved in national and international not-for-profit organizations that encourage ethical medical writing practices. The author has no relationships with any pharmaceutical company that might have an interest in the submitted work and has no other relationships that could be construed as a competing interest.

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DATA SHARING

All data associated with the study are fully available to the author.

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FIGURE LEGENDS

Figure 1. Selection of publications reporting a numerical estimate of the prevalence of ghostwriting. Electronic databases were searched on 23 May 2013.

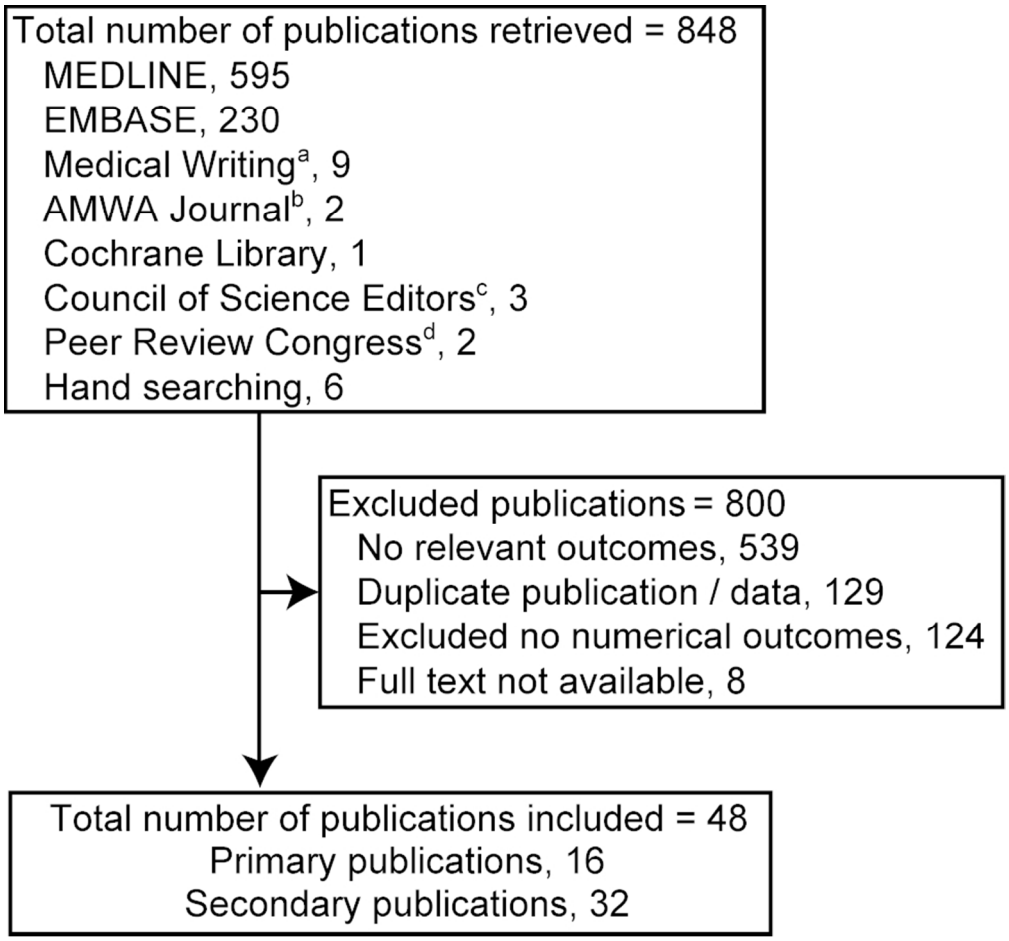
[a] Journal of the European Medical Writers Association, formerly known as The Write Stuff.

[b] Journal of the American Medical Writers Association.

[c] Council of Science Editors Annual Meetings.

[d] International Congress on Peer Review and Biomedical Publication.

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[6] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.



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Electronic databases were searched on 23 May 2013.
a Journal of the European Medical Writers Association, formerly known as The Write Stuff.
b Journal of the American Medical Writers Association.
c Council of Science Editors Annual Meetings.
d International Congress on Peer Review and Biomedical Publication.

82x76mm (300 x 300 DPI)

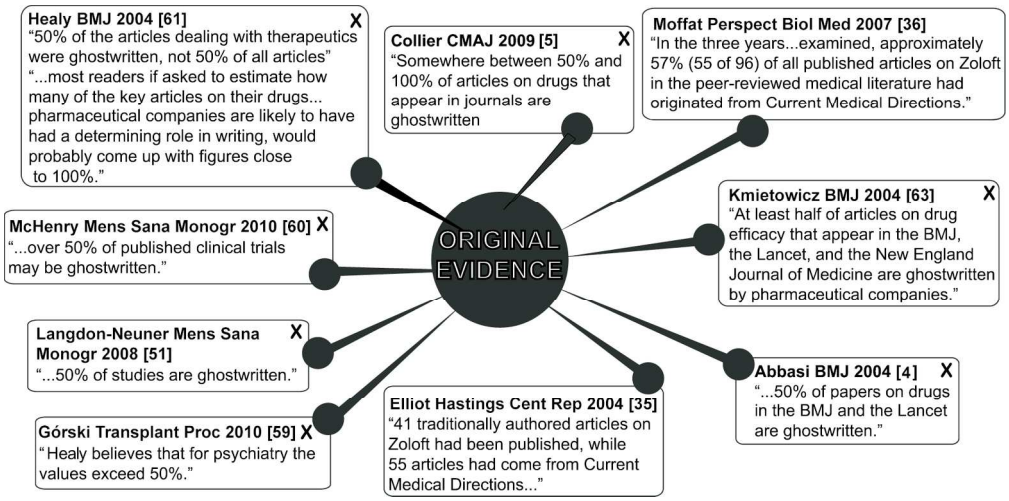

Figure 2. Case study of original versus cited evidence of ghostwriting prevalence. Contrast between original evidence on the prevalence of possible ghostwriting in sertraline publications from 1998 to 2000 and the subsequent citations of this evidence. Source of the original published evidence: Healy and Cattell[6] showed that 55 of 96 (57%) articles published on sertraline from 1998 to 2000 were coordinated through a medical communications company and concluded that these data provided information on the "possible extent of ghostwriting based on a single drug". Inaccurate reporting from the cited source is marked with a cross.
170x83mm (300 x 300 DPI)

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	p. 1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	pp. 2/3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	p. 5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	p. 6
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	A review protocol was prepared that prespecified the primary and secondary objectives of the literature review, eligibility criteria, data to be collected, and primary and secondary outcome measures. The review protocol was not registered in a public registry but is available upon request.
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	p. 7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	p. 6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	p. 6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	pp. 6/7
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	p. 7

PRISMA 2009 Checklist

Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	p. 7
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	Risk of bias was not conducted formally because data were not synthesised and because of the various study types retrieved. Given the various study designs and methods used across the retrieved studies, the quality of the study methods were assessed on a study-by-study basis. The key factors considered were the study design, definition of ghostwriting used, and study population. The factors considered when determining the quality of the cross-sectional surveys are described in the Methods section (p. 7).
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	Not applicable because study data were not synthesised
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	Not applicable because study data were not synthesised.

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	Risk of bias across studies was not conducted formally because data were not synthesised and because of the considerable variation in study designs, populations, and definitions of ghostwriting used in the retrieved publications.
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	No additional analyses were conducted
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	8, Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	8-11, 15, Tables 1-3
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Risk of bias was not conducted formally because data were not synthesised. All studies retrieved were limited by the observational nature of the data collection. The quality of individual components of each study are listed in Table 1

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			and are summarised in the results section (Publication Characteristics) (pp. 8/9).
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Tables 1-3
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	Not applicable because study data were not synthesised
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	Not applicable because study data were not synthesised and because of the considerable variation in study designs, populations, and definitions of ghostwriting used in the retrieved publications.
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	No additional analyses were conducted
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	20-24
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	20-22
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	22-24
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	No funding was received to conduct this study

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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