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The Impact of Fraudulent Research: A Citation Perspective on the Breuning Case

Number 12

In May 1989 the First International Congress on Peer Review in Biomedical Publication was held in Chicago, Illinois. The title of the three-day congress, sponsored by the American Medical Association, was "Guarding the Guardians: Research on Peer Review."¹ Examining peer review from many viewpoints, the conference discussions included the history, evolution, and current status of the peer-review process, the existence of publication bias, and the accuracy of quotations and references. Not surprisingly, given the media attention the issue has received, scientific fraud was one of the major topics at the congress.

I was invited to participate in the conference by Drummond Rennie, deputy editor (west), JAMA, the Journal of the American Medical Association. Drummond knew of ISI[®]'s preliminary analysis of citations to the work of John R. Darsee, then a researcher at the Harvard Medical School Cardiac Research Laboratory, Boston, Massachusetts. Darsee was caught fabricating data at the laboratory in 1981 in an animal experiment involving radioactive tracers and heart tissue.² Drummond suggested we perform a similar analysis on the work of Stephen E. Breuning, then a psychologist at the University of Pittsburgh, Pennsylvania, the first researcher to be tried and convicted of fraud.^{3,4} The Breuning case is especially interesting to clinicians because it involved research on patients and influenced at least one state's policies on care for the mentally retarded.

Scientific fraud⁵⁻⁷ and peer review⁸ have been discussed extensively in *Current Con*-

tents[®]. Drummond's invitation gave me an opportunity to pursue a basic question that has interested me for some time: What impact does published fraudulent work have on other investigators who use the literature? Have they been led astray by fictitious data, wasting their time and resources pursuing false leads? Or have they realized, even before the fraud is exposed, that the published data were suspect and avoided citing them?

In the Breuning case, at least, my colleague Alfred Welljams-Dorof and I found some interesting results. For example, of the 20 Breuning publications examined, 11 received at least 10 citations. Judging by total citations alone, one might conclude that Breuning's work had high impact-only 7 percent of the 30 million cited items in the 1955-1987 Science Citation Index[®] (SCI[®]) files were cited 10 or more times. However, further analysis of these citations diminishes the putative impact of Breuning's research: 40 percent were self-citations and about 33 percent of non-self-citations disagreed with Breuning's findings and/or methods. Also, after Breuning's fraud was exposed in 1986, citations to his work declined sharply-indicating that researchers seem to shun work that is known or even suspected to be falsified. These and other results were presented at the peer-review congress and published in a special JAMA issue (March 9, 1990) that carries reports from the conference.9 The article is reprinted here.10

Of course, a single case study on a specialized topic cannot completely answer all the questions raised by the issue of scientif-

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ic fraud and its impact on the literature. For example, what happens to the legitimate research of an author who is exposed for having published fraudulent work? Do other researchers shun only the fraudulent papers or are *all* of the author's papers stigmatized? If this "stigma effect" is documented by citation analysis, does it also extend to coauthors of fraudulent work who were unaware that falsified results were being reported? Is their independent work stigmatized as well? It would be a complicated and difficult task to test these ideas, but it would be worth the effort and might make an excellent thesis topic for information-science scholars.

As the debate on scientific fraud continues, a point made in the following reprint should be remembered: citation indexes can be used to help ensure that fraudulent or erroneous research results are not relied upon in ignorance of relevant corrections or retractions. In the SCI all correction notes are indexed—most of these are the usual "errata," but some are also formal retractions. In addition to correction notes, other significant publications-that is, research articles and reviews, editorials, letters to the editor-may also contain important caveats about previously published work. If researchers, editors, and reviewers used citation indexes routinely to check references. papers containing erroneous, obsolete, or falsified data would be more easily identified, thereby preventing or at least limiting their use. Editors can easily make such routine checks in a cost-effective manner by using the compact-disc edition of the SCI. which now covers the literature from 1980 onwards, 11

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My thanks to Peter Pesavento for his help in the preparation of this essay.

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The Impact of Fraudulent Research on the Scientific Literature

The Stephen E. Breuning Case

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The goal of this study was to determine the research impact of scientific fraud through citation analysis of 20 Breuning publications, using the 1980 to 1988 Science Citation Index[®] and Social Sciences Citation Index[®]. These publications received 200 citations, of which 80 (40.0%) were self-citations by Breuning or his coauthors. Tracked over time, non-self-citations declined sharply in 1986 and later years, coinciding with disclosure of Breuning's fraud. The data indicated that, in this case, researchers effectively shunned work known to be or even suspected of being falsified. Unique citation contexts (101) were examined to see how citing authors used Breuning's work: 33 were negative (disagreed with findings/methods), 10 positive (agreed), and 58 neutral (no valuation). Also, 63 were led to negative conclusions. These data diminish the apparent impact of Breuning's work suggested by total citations alone.

JOURNAL editors are currently debating what, if anything, they can and should do to deal effectively with falsified research that escapes the traditional quality-control filter of peer review. One proposal would involve conducting data audits of submitted papers to prevent publication of fraudulent work.1-4 For clinical research, data audits may be a relatively straightforward task. Hospital records could easily be checked to verify that patients were indeed studied and protocols were followed as reported. But data audits of basic biomedical research, particularly in newly emerging specialties, may prove to be a complex and ambiguous undertaking. It may be difficult, if not impossible, to find auditors who are willing or able to make the subtle interpretations of original data needed to judge whether an author's analysis is sound, unwittingly erroneous, or intentionally fraudulent.

Another proposal would focus on *correcting* the scientific record by strengthening existing mechanisms that alert researchers to published studies that subsequently are exposed as fraudulent.⁵⁻⁷ This would simply involve the prompt and prominent publication of explicit correction or retraction notices. However, journal editors are reluctant to print retractions or even report the findings of independent investigations because they fear legal action by discredited authors or their coauthors. To overcome this obstacle, Congress is considering new laws that would grant scien-

tific journals immunity to publish retractions in good faith, regardless of whether all authors of a discredited study consent to it.

The current debate on scientific fraud continues to focus on the small but growing number of studies that report falsified research. Little attention has been paid to the question of what impact these studies have on research. That is, how frequently were they cited, and, just as important, how were they used by citing authors?

The present study is a first effort to answer this question through a citation analysis of publications by Stephen E. Breuning, who in 1988 was prosecuted and convicted in federal court of scientific fraud (*New York Times*. May 24, 1987:A16).⁸⁻¹⁰ Additional citation studies of other known cases of scientific fraud are needed to assess systematically their general impact on research. Of course, fraud involving clinical research has potentially profound consequences for patient care and should be carefully examined by the medical community.

To summarize briefly the case examined here, Breuning published a number of studies from 1980 to 1984 on the use of drugs to control hyperactive retarded children while at the Coldwater (Mich) Regional Center for Developmental Disabilities and the University of Pittsburgh (Pa). Purportedly based on hundreds of human subjects, the studies claimed that stimulant drugs were more effective and had fewer side effects than tranquilizers, the traditional drug therapy for retarded children. His findings supported a controversial theory that less immediate drug intervention, combined with other nondrug treatments, would be most beneficial. Experts in the field have claimed that Breuning's work was influential and even led some states, notably Connecticut, to change their policies on treating retarded children. But an investigation by the National Institute of Mental Health (NIMH), initiated after a colleague challenged Breuning's work, found that he had "knowingly, willfully, and repeatedly engaged in misleading and deceptive practices in reporting results."10 The NIMH report, released in May 1987, concluded that "None of the described studies of psychopharmacologic treatment had been carried out," "Only a few of the experimental subjects...were ever studied," and "The complex designs and rigorous methods reported were not employed."10 The NIMH referred the case to the US attorney's office in Maryland for criminal prosecution, and, in September 1988, Breuning pleaded guilty to two counts of making false statements on federal grant applications. He was later sentenced to serve 60 days in a halfway house, 250 hours of community service, and 5 years of probation.

METHODS

The files of the Science Citation Index[®] and Social Sciences Citation Index[®] from 1980 to 1988 were used to identify 23 cited Breuning publications. Three of these were deleted from this study. One was a book he coedited, which was cited 14 times without reference to specific chapters and pages.¹¹ Another was a book chapter reported to be in press but never subsequently published (1 citation). The last was a 1984 methods article (8 citations) describing a rating scale for tardive dyskinesia.¹²

Citation counts were tabulated for each of the 20 cited articles included in the study. These data were divided into two categories: self-citations and non-self-citations. *Self-citations* refer to citing articles that were authored by Breuning or one of the coauthors of the cited work. The annual distribution of each category of citations was also determined.

In addition, a citation context analysis was conducted to determine how citing authors used Breuning's work. This involved retrieving complete copies of 65 citing articles (excluding selfciting papers) and identifying in each text where the authors cited Breuning's work and how they referred to it. A total of 183 citation contexts were identified and categorized. The first category distinguished between serial and unique citation contexts. In serial citation contexts, Breuning's work was cited in combination with publications by other authors. That is, his work was embedded with that of a group of other researchers, all reporting similar claims, findings, statistics, or conclusions. These group citations were excluded from further analysis, which focused on the unique citation contexts that referred only to Breuning studies.

Second, the unique citation contexts were categorized as positive, negative, or neutral. *Positive citations* were defined as references that indicated agreement with Breuning's methods, findings, or conclusions. *Negative citations* indicated disagreement. Neutral citation contexts made no explicit valuation of his work but simply stated what he had reported.

Finally, the unique citation contexts were categorized as material or inconsequential. Material citation contexts indicated that Breuning's work influenced the interpretation of data and/or the formation of conclusions in a citing author's article. Inconsequential citation contexts are those in which no such influence was observed.

RESULTS

Table 1 lists the 20 Breuning publications included in this study, ranked by the number of times each was cited from 1981 to 1988 in the *Science Citation Index* and *Social Sciences Citation Index*. Fifteen are journal articles and 5 are book chapters.

These publications were cited 200 times in 86 articles. Of these, 80 (40.0%) were self-citations by Breuning or his coauthors and 120 (60.0%) were non-self-citations.

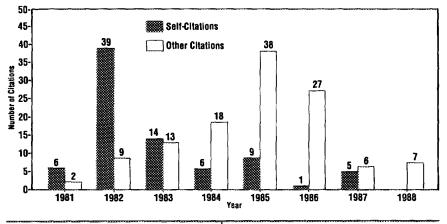
The Figure presents a graph of the annual distribution of self-citations and non-self-citations. Two distinct patterns are obvious. Self-citations peak rapidly at 39 in 1982, decline noticeably to 14 in 1983, and continue downward thereafter. This trend results primarily from the decreasing number of articles by Breuning over time: as he published less, he had fewer opportunities to cite himself. For example, in 1981 nine Breuning articles were indexed in the Science Citation Index and Social Sciences Citation Index, in which he cited himself six times. In 1982, six articles containing 32 self-citations were indexed. In 1983, three articles with 11 self-citations were indexed. Two Breuning articles indexed in 1984 contained 2 self-citations, and two in 1985 included 7 self-citations.

Non-self-citations show a more gradual but steady increase, peaking at 38 in 1985 and declin-

Table 1Cited Publications by Stephen E. Breunin	, 1980 Through 1988, in the Science Citation Index [®] and
Social Sciences Citation Index [®] .	

No. of Citations	Publication
26	Breuning SE, O'Neill MJ, Ferguson DG. Comparison of psychotropic drug, response cost, and psychotropic drug plus response cost procedures for controlling institutionalized mentally retarded persons. <i>Appl Res Ment Retard.</i> 1980;1:253-268. Retracted.
25	Breuning SE, Davidson NA. Effects of psychotropic drugs on intelligence test performance of institutionalized mentally retarded adults. Am J Ment Defic. 1981;85:575-579.
18	Davis VJ, Cullari S, Breuning SE. Drug use in community foster group homes. In: Breuning SE, Poling AD, eds. Drugs and Mental Retardation. Springfield, Ill: Charles C Thomas Publisher; 1982:359-76.
15	Breuning SE. An applied dose-response curve of thioridazine with the mentally retarded: aggressive, self-stimulatory, intellectual, and workshop behaviors—a preliminary report. <i>Psychopharmacol Bull.</i> 1982;18:57-59.
15	Breuning SE, Davis VJ, Matson JL, Ferguson DG. Effects of thioridazine and withdrawal dyskinesias on workshop performance of mentally retarded young adults. <i>Am J Psychiatry</i> . 1982;139:1447-1454. Retracted.
13	Breuning SE, Ferguson DG, Davidson NA, Poling AD. Effects of thioridazine on the intellectual performance of mentally retarded drug responders and nonresponders. <i>Arch Gen Psychiatry</i> . 1983;40:309-313.
13	Davis VJ, Poling AD, Wysocki T, Breuning SE. Effects of phenytoin withdrawal on matching-to- sample and workshop performance of mentally retarded persons. J Nerv Ment Dis. 1981;169:718-725.
12	Breuning SE, Ferguson DG, Cullari S. Analysis of single- and double-blind procedures, maintenance of placebo effects, and drug-induced dyskinesias with mentally retarded persons. <i>Appl Res Ment Retard.</i> 1980;1:175-192. Retracted.
12	Breuning SE, Poling AD. Pharmacotherapy with the mentally retarded. In: Matson JL, Barrett RP, eds. <i>Psychopathology of the Mentally Retarded</i> . New York, NY: Grune & Stratton; 1982:195-251.
12	Wysocki T, Fuqua W, Davis VJ, Breuning SE. Effects of thioridazine (Mellaril) on titrating delayed matching-to-sample performance of mentally retarded adults. <i>Am J Ment Defic.</i> 1981;85:539-47.
10	Breuning SE, Davis VJ, Poling AD. Pharmacotherapy with the mentally retarded: implications for clinical pathologists. <i>Clin Psych Rev.</i> 1982;2:79-114.
7	Gualtieri CT, Breuning SE, Schroeder SR, Quade D. Tardive-dyskinesia in mentally retarded children, adolescents, and young adults: North Carolina and Michigan studies. <i>Psychopharmacol Bull.</i> 1982;18:62-65.
5	Ferguson DG, Breuning SE. Antipsychotic and antianxiety drugs. In: Breuning SE, Poling AD, eds. Drugs and Mental Retardation. Springfield, Ill: Charles C Thomas Publisher; 1982:168-214.
5	Ferguson DG, Cullari S, Davidson NA, Breuning SE. Effects of data-based interdisciplinary medication reviews on the prevalence and pattern of neuroleptic drug use with institutionalized mentally retarded persons. <i>Educ Train Ment Retard</i> . 1982;17:103-108.
4	Poling A, Breuning SE. Effects of methylphenidate on the fixed-ratio performance of mentally retarded children. <i>Pharmacol Biochem Behav.</i> 1983;18:541-544.
2	Breuning SE, Ferguson DG, Cullari S. Analysis of single-blind, double-blind procedures, maintenance of placebo effects, and drug-induced dyskinesia with mentally retarded persons: a brief report. <i>Psychopharmacol Bull.</i> 1981;17:122-123.
2	Matson JL, Breuning SE. A review and analysis of applied research in mental retardation: 1975-1980. Appl Res Ment Retard. 1982;3:185-189.
2	Poling AD, Breuning SE. Overview of mental retardation. In: Breuning SE, Poling AD, eds. Drugs and Mental Retardation. Springfield, Ill: Charles C Thomas Publisher; 1982:3-65.
1	Breuning SE, Davis VJ. Reinforcement effects on the intelligence test performance of institutionalized retarded adults: behavioral analysis, directional control, and implications for habilitation. <i>Appl Res Ment Retard.</i> 1981;2:307-322.
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Annual distribution of citations to Breuning publications, 1981 through 1988. Shaded bars indicate self-citations; and open bars, non-self-citations (data from the Science Citation Index[®] and Social Sciences Citation Index[®], Institute for Scientific Information[®], Pa).



ing to 27 in 1986, 6 in 1987, and 7 in 1988. Four of the 18 citations in 1987 and 1988 were retractions or corrections published as editorials or letters to the editor. This decline coincides with the publication of a critical review of Breuning's work in the September 1986 issue of the Journal of Mental Deficiency Research.¹³ The review, by M. G. Aman, PhD, of the University of Auckland and N. N. Singh, PhD, of the University of Canterbury, New Zealand, highlighted various methodological shortcomings of Breuning's work, noted the lack of corroborating data from other researchers, pointed to other studies that showed opposite results, and concluded that it "does not substantially advance our level of knowledge, despite what initially appeared to be an unusual level of methodological elegance in the way the studies were conducted."13

In a 1987 guest editorial in the same journal, Aman¹⁴ said the review was initiated in 1985 in part by his knowledge that the NIMH was investigating Breuning's work. If researchers in New Zealand were aware of the NIMH inquiry in 1985, it is reasonable to assume that other investigators in the field also knew of the fraud allegations against Breuning and began to avoid his work. In any event, the NIMH investigation was publicly disclosed in the December 19, 1986, issue of *Science*.¹⁵

As noted earlier, 183 citation contexts in 65 citing articles were examined to determine how Breuning's work was used by the citing authors. Of these, 82 (44.8%) were serial citations and 101 (55.2%) were unique citations. Results of an analysis of these unique citation contexts are summarized in Table 2.

The majority of the unique citation contexts (58,or 57.4%) were neutral, in which the citing author simply reported some aspect of Breuning's work. However, 33 (32.7%) were negative, indicating disagreement with Breuning's findings and/or criticism of his methods. Only 10 (9.9%) of the citation contexts were positive.

Categorized another way, the majority of the unique citation contexts (63, or 62.4%) were inconsequential, indicating that Breuning's work had no apparent influence on the interpretation of data and/or conclusions reported in the citing author's study. Thirty-eight (37.6%) of the citation contexts were material, and they were contained in 15 (23.1%) of the 65 articles examined.

However, it should be stressed that the critical review by Aman and Singh accounted for 21 (55.3%) of the material citation contexts. Breuning's work did indeed influence the conclusions formed by the authors, but in an ironic way. As noted earlier, they concluded that his work "does not substantially advance our level of knowledge."¹³

COMMENT

The most interesting observation to emerge from this single case study is that the scientific literature seems to purge itself of articles that are known or even suspected to be fraudulent. The annual distribution of non-self-citations indicates

Table 2	Results of Analysis o	f 101	Unique Breuning
Citation	Contexts*		

	Neutral	Negative	Positive	Total
Inconsequential	45	13	5	63
Material	13	20	5	38
Total	58	33	10	101

*See "Methods" section for definition of categories.

that authors shun falsified research once it is publicly exposed.

Also, the frequency of citations to Breuning's work seems to indicate that it was influential in the field. Table 1 shows that 11 Breuning publications were cited between 10 and 26 times. In the 1955 to 1987 Science Citation Index files of 30 million cited items, only 7% achieved this level of citation. However, closer examination of the type and context of Breuning citations diminishes the apparent influence of his work. First, a high percentage (40.0%) were self-citations. Second, a high percentage of non-self-citations (32.7%) disagreed with Breuning's findings or criticized his methods. Last, while a significant proportion (37.6%) of Breuning citations were material, most of these (55.3%) led to a negative conclusion about his work.

In conclusion, this study suggests the potential value of citation indexes for limiting the spread of falsified research. Citation indexes can make readers aware of explicit retraction notices, provided these are published in a suitable form for proper indexing. But such notices are rare. Just as important, citation indexes can lead readers to reviews, editorials, letters to the editor, and comments that may contain important caveats about previously published work.^{16,17}

In fact, the ability to eliminate the uncritical citation of fraudulent, incomplete, or obsolete data was a primary reason for developing citation indexes. A 1955 article that first described the uses of citation indexes in science¹⁸ opened with the following quotation of P. Thomasson and J. C. Stanley, of The Johns Hopkins University, Baltimore, Md, which is still relevant today: The uncritical citation of disputed data by a writer, whether it be deliberate or not, is a serious matter. Of course, knowingly propagandizing unsubstantiated claims is particularly abhorrent, but just as many naive students may be swayed by unfounded assertions presented by a writer who is unaware of the criticisms. Buried in scholarly journals, critical notes are increasingly likely to be overlooked with the passage of time, while the studies to which they pertain, having been reported more widely, are apt to be rediscovered.¹⁹

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