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How Statistical Expertise Is Used in Medical Research

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Context Investigation of the nature and frequency of statistician involvement in medical research and its relation to the final editorial decision.

Methods Authors of original research articles who submitted to *BMJ* and *Annals of Internal Medicine* from May through August 2001 were sent a short questionnaire at the time of manuscript submission. Authors were asked if they received assistance from a person with statistical expertise, the nature of any such contribution, and reasons why, if no statistical input was received.

Results The response rate was 75% (704/943); methodological input was reported for 514 (73%) of these papers. In 435 papers (85%), such input was provided by biostatisticians or epidemiologists and, if deemed significant, was typically associated with authorship. A total of 33 of 122 methodologists (27%) whose main contribution started at the analysis stage received neither acknowledgment nor authorship. Research without methodological assistance was more likely to be rejected without review (71% vs 57%; χ^2=10.6; P=.001) and possibly less likely to be accepted for publication (7% vs 11%; χ^2=2.37; P=.12).

Conclusions Statistical input to medical research is widely recommended but inconsistently obtained. Individuals providing such expertise are often not involved until the analysis of data and many go unrecognized by either authorship or acknowledgment.

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STATISTICAL CONTENT AND COMplexity of medical research has increased steadily over recent decades.[1-3] Although there is considerable evidence that methodological errors are common in articles in medical journals,[4-6] much published research does not have substantive contribution from a statistician. Anecdote suggests that many physicians have difficulty getting expert advice or involvement in their research and statisticians are often brought in only at the analysis stage or later.

In 1949, Luykx[7] wrote, "It is now almost inconceivable that a study of any dimensions, in medical science, can be planned without the advice of a statistician." He clearly appreciated that the most important contribution of the statistician to medical research is at the design stage. We are unaware of any study of the degree to which persons with quantitative expertise are involved in medical research. We report a survey of authors submitting to 2 major medical journals to investigate the nature and frequency of such

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involvement and its relation to the final editorial decision.

METHODS

We sent a short questionnaire to authors of research articles immediately after manuscript submission. Eligible papers were all those submitted to the *BMJ* and *Annals of Internal Medicine* from May through August 2001. The only papers excluded were those not reporting original research on humans. Questionnaires were sent by mail to *BMJ* authors and electronically to *Annals* authors.

Recognizing that several kinds of researchers have statistical expertise, we asked authors the following question: "Was a statistical consultant (or someone with graduate training or a qualification in quantitative research methods) involved at any stage in your research?" If they answered yes, authors were asked about the qualifications (degrees) of the person providing that contribution and whether they were employed as a biostatistician, epidemiologist, or other. For analysis, some answers of other (eg, statistician, clinical epidemiologist) have been reclassified into one of the other categories.

We also asked about the extent of involvement of the methodologist at each stage of the project (none or minimal, moderate, or significant), how they were credited in the paper (author, acknowledged, or neither), and whether they received payment or salary support for the particular project. If there were 2 such individuals, they were asked to supply information for the senior person or the one whose contributions most affected the paper. Where there had been no expert statistical input, authors were asked to indicate reasons from a list of possibilities, whether they would have liked to have had a statistical consultant involved in their project, and whether they had previously worked with a statistician.

Authors were told that the processing of their paper by the journal would not be affected by their answers or whether they responded. The cover letter was signed by statistical advisors to the 2 journals (D.G.A. and S.N.G.) and noted that the study was supported by the editors of the journals, Richard Smith (*BMJ*) and Frank Davidoff (*Annals*). Authors were asked to send their completed questionnaire directly to the relevant statistician (D.G.A. or S.N.G.) and their replies were blinded to any editorial staff at the journals. For the *BMJ*, reminders were sent after 3 to 5 weeks only to nonresponding authors who had provided an e-mail address (around 90%). For the *Annals*, a reminder was sent by e-mail to all nonrespondents after 1 week.

Each abstract was read to classify papers according to their study design as a randomized controlled trial (RCT), systematic review (meta-analysis), observational study, economic study, or other (at the *BMJ*, the whole paper was examined in cases of uncertainty). The category "observational" included epidemiologic studies of various designs, nonrandomized clinical cohort studies, prognostic studies, qualitative studies, and surveys. From each journal's editorial database, each paper was classified as rejected without review, rejected after review, or accepted for publication. Proportions were compared using χ^2 tests, with 1 degree of freedom unless stated otherwise. Stata statistical software release 6.0 (Stata-corp, College Station, Tex) was used for all analyses.

Table 1. Methodological Input in Relation to Study Design

	Methodologist, No./Total (%)		
Design	Biostatistician	Epidemiologist	Other
Randomized controlled trial	43/65 (66)	15/65 (23)	7/65 (11)
Systematic review	18/34 (53)	14/34 (41)	2/34 (6)
Observational	197/385 (51)	127/385 (33)	61/385 (16)
Economic	8/14 (57)	3/14 (21)	3/14 (21)
Other	7/16 (44)	3/16 (19)	6/16 (38)
Total	**273/514** (53)	**162/514** (32)	**79/514** (15)

Table 2. Proportion of Papers With a Methodologist Involved in Which That Individual Made a Moderate or Significant Contribution at Each Stage in Relation to Study Design*

	No./Total (%)				
Design	Formulating the Study Question	Developing the Study Design	Conducting the Research	Analyzing the Data	Writing the Paper
Randomized controlled trial	29/65 (45)	47/65 (72)	17/65 (26)	62/65 (95)	42/65 (65)
Systematic review	20/33 (61)	26/33 (79)	19/33 (58)	31/33 (94)	23/33 (70)
Observational	198/384 (52)	248/384 (65)	174/384 (45)	365/384 (95)	262/384 (68)
Economic/other	18/29 (62)	22/29 (76)	14/29 (48)	26/29 (90)	20/29 (69)
Total	**265/511** (52)	**343/511** (67)	**224/511** (44)	**484/511** (95)	**347/511** (68)

*Three nonresponders for this section.

RESULTS

Responses were received from 75% of authors (704/943). The response rate from *BMJ* authors was higher than from *Annals* authors (585/741 [79%] vs 119/202 [59%]). The included studies comprised 103 RCTs (11%), 52 systematic reviews (6%), 730 observational studies (78%), 27 economic studies (3%), and 31 other (3%). The distribution was similar for both journals apart from more economic evaluations at *Annals* (8% vs 1.5%). The response rate was broadly similar in all categories.

Input from a methodologist was reported for 514 of 704 papers (73%): 273 were biostatisticians (53%), 162 were epidemiologists (32%), and 79 were neither (15%). Reported qualifications varied greatly but the majority of biostatisticians and epidemiologists had a PhD or equivalent degree and only a

few did not have a masters-level qualification. Biostatisticians were most likely to be involved in RCTs, epidemiologists in systematic reviews, and other methodologists in economic studies and other designs (TABLE 1).

The input from the methodologist was most often classified as moderate or significant for analyzing the data (95%), developing the study design (67%), and writing the paper (68%) (TABLE 2). The first moderate or significant contribution from the methodologist occurred at the analysis stage for 30% of biostatisticians (82/270), 16% of epidemiologists (26/162), and 20% of others (16/79; $\chi^2_2 = 12.1$; $P = .002$). Biostatisticians were more likely to make their first important contribution before the analysis stage for RCTs (35/43 [81%]) than for other study designs (153/227 [68%]; $\chi^2 = 3.35$; $P = .07$).

TABLE 3 shows that biostatisticians were much less likely to be authors and more likely not even to be acknowledged than epidemiologists or other methodologists (2 authors reported that a biostatistician had declined authorship). Among those who were reported to have made a significant contribution to at least 1 of the 5 study stages, 22% of biostatisticians were not authors compared with 4% of epidemiologists and 10% of other methodologists ($\chi^2_2 = 21.7$; $P < .001$). One in 7 of these biostatisticians were neither authors nor acknowledged. For those who had made only a minimal contribution prior to analysis, authorship was less common, and 27% of these methodologists were not even acknowledged.

Specific payment for the project described in the paper was reported for 37% of biostatisticians, 20% of epidemiologists, and 29% of others. Authorship was reported for 78% of methodologists who received payment and 74% who did not. Payment was more common for RCTs (30/65 [46%]) than for other types of study (122/438 [28%]).

For 190 papers (27%), there had not been any expert methodological input. The most common reason given (61%) was "we felt we had the necessary skills within the research team." More seriously, 13% of these authors noted that no statistical consultant was available. Many of these authors (64/180 [36%]) would have liked a statistical consultant involved in their project, regardless of whether they felt they had the necessary skills (37%) or not (31%). Most (147/183 [80%]) had previously worked with a statistician.

TABLE 4 shows the editorial outcome of the papers. A total of 60% of the papers were rejected without going to external peer review (*BMJ*, 65%; *Annals*, 42%). Papers with no methodologist were more likely to be rejected without going to peer review compared with papers with methodological input (71% vs 57%; $\chi^2 = 10.6$; $P = .001$). Rejection without peer review was less common for RCTs and systematic reviews (48/118 [41%]) than for other study designs (379/586 [65%]; $\chi^2 = 23.7$; $P < .001$). Papers with a methodologist were more likely to be accepted for publication (55/514 [11%]) than those without (13/190 [7%]; $\chi^2 = 2.37$; $P = .12$).

COMMENT

We believe this study represents the first attempt to survey authors to ascertain the use and nature of expert methodological assistance in the development and analysis of clinical research submitted for publication. We found that such assistance was used for about 75% of the papers, more than 80% of such input was provided by biostatisticians or epidemi-

Table 3. Frequency of Recognition of Contribution of Methodologist as Author or Acknowledged

	No. (%)			
	Biostatistician	Epidemiologist	Other	Total
All Papers				
Author	176 (65)	141 (88)	65 (82)	**382** (75)
Acknowledgment	40 (15)	6 (4)	8 (10)	**54** (11)
Neither	53 (20)	13 (8)	6 (8)	**72** (14)
Total	**269** (100)	**160** (100)	**79** (100)	**508** (100)*
Papers for Which the Methodologist Had Made a Significant Contribution at Some Stage				
Author	158 (78)	128 (96)	61 (90)	**347** (86)
Acknowledgment	17 (8)	2 (1)	7 (10)	**26** (6)
Neither	28 (14%)	4 (3)	0	**32** (8)
Total	**203** (100)	**134** (100)	**68** (100)	**405** (100)
Papers for Which the Methodologist Made First Moderate or Significant Contribution at the Analysis Stage				
Author	37 (46)	14 (56)	8 (50)	**59** (48)
Acknowledgment	20 (25)	5 (20)	5 (31)	**30** (25)
Neither	24 (30)	6 (24)	3 (19)	**33** (27)
Total	**81** (100)	**25** (100)	**16** (100)	**122** (100)

*Six outcomes were not known.

Table 4. Editorial Outcome in Relation to Methodological Input

	No. (%)				
	Methodologist				
Editorial Decision	Biostatistician	Epidemiologist	Other	No Methodologist	Total
Rejected without peer review	152 (56)	92 (57)	49 (62)	134 (71)	**427** (61)
Rejected after peer review	95 (35)	46 (28)	25 (32)	43 (23)	**209** (30)
Accepted	22 (8)	21 (13)	4 (5)	12 (6)	**59** (8)
Peer reviewed, final outcome not yet known*	4 (1)	3 (2)	1 (1)	1 (0)	**9** (1)
Total	**273** (100)	**162** (100)	**79** (100)	**190** (100)	**704** (100)

*Very likely to be accepted.

ologists, research not using this assistance was more likely to be rejected, and if such input was deemed significant, it was typically associated with authorship. However, 27% of methodologists whose contribution was moderate or significant but started at the analysis phase received neither acknowledgment nor authorship.

Because the survey was conducted of submitted, not published papers, it represents a more generalizable snapshot of the use of statistical expertise in clinical research at present than any study of published research. Although the study was conducted at 2 major medical journals, more than 85% of the papers were rejected and many will be published elsewhere.

The importance of having persons with quantitative expertise as part of the research team has been stressed for decades, but this survey provides evidence that their potential contributions may still not be fully appreciated. Biostatisticians and epidemiologists have expertise not just in analysis but in the design of studies and it is at that phase that their input is likely to be most valuable. Expert analysis cannot salvage poorly designed research, yet it is clear that many authors do not use methodological assistance for study design. In papers with methodologists involved, 24% of those individuals (and 30% of statisticians) had little input before data analysis. It is perhaps not coincidental that in such papers nearly a third of the biostatisticians received no acknowledgment of any involvement. In papers without methodologists, 18% of authors gave the lack of quantitative methods as a reason.

Empirical data on the value of expert statistical help in the conduct of medical research have been indirect, mainly confined to analyses of statistical errors in published papers.[4-6] Two recent studies found that the quality of published controlled clinical trials[8] and the reporting of statistical adjustment procedures[9] were better in papers with a methodologist (biostatistician or epidemiologist) among the authors. The present study shows that authorship is an imperfect indicator of a methodologist's involvement. Indeed, in both of these studies,[8,9] a methodologist was apparently involved in 38% of papers compared with 73% in the present study. In addition, the contribution of methodologists may not always be apparent just from the presence or absence of gross errors. With the use of increasingly complex statistical technologies, such collaborators are often necessary for an accurate assessment of whether the assumptions of such methods have been met and an accurate representation of the attendant uncertainty in the conclusions. A statistical collaborator can be essential in crafting appropriately nuanced language in the discussion section, subtleties that are difficult to capture in a standardized instrument of report quality.

Our study must be regarded as mainly descriptive, as there are many aspects that make inferences difficult. First, the response rate of 75%, while reasonable, probably selected for authors who were more likely to use statistical help. Second, authors self-select when they choose a journal and this group of manuscripts may represent a level of research more likely to use both quantitative methods and collaborators with related expertise. Both *Annals* and *BMJ* may be known by many authors as having stronger statistical reviewing policies than most other journals.[10] Third, we did not assess the quality of the submitted papers nor did we independently assess the extent to which specialized statistical expertise was necessary in these papers; thus, it is not possible to posit the right proportion of papers that should have reported having statistical assistance. Finally, the studies most likely to be accepted at either of these journals, RCTs and large cohort studies, are also those most likely to use expert quantitative help. Thus, the relationship of acceptance rates to statistical assistance is undoubtedly strongly confounded by study type. Stratification by design may have reduced but not eliminated the problem. Nevertheless, this study provides a picture of the norms and practices of this aspect of the medical research enterprise in 2001 and identifies several areas for possible exploration and improvement in the future.

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