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Publication Bias in Kienbock's Disease: A Systematic Review

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Abstract

Purpose—Kienbock's disease is considered a “rare disease” and currently affects less than 200,000 people in the U.S. Given the inherent challenges associated with researching rare diseases, the intense effort in hand surgery to treat this uncommon disorder may be susceptible to publication bias in which positive outcomes are preferentially published. The specific aim of this project is to conduct a systematic review of the literature with the hypothesis that publication bias is present for the treatment of Kienbock's disease.

Methods—We conducted a systematic review of all available abstracts associated with published manuscripts (English and non-English) and abstracts accepted to the 1992-2004 American Society for Surgery of the Hand (ASSH) annual meetings. Data collection included various study characteristics, direction of outcome (positive, neutral/negative), complication rates, mean follow-up time, time to publication, and length of patient enrollment.

Results—Our study included 175 (124 English, 51 non-English) published Kienbock's manuscripts and 14 Kienbock's abstracts from the 1992-2004 annual ASSH meetings. Abstracts from published manuscripts were associated with a 53% positive outcome rate, which is lower than the 74% positive outcome rate found among other surgical disorders. Over the past 40 years, studies have become more positive (36% to 68%, $p = 0.007$) and are more likely to incorporate statistical analysis testing (0% to 55%, $p < 0.001$). Of the 14 abstracts accepted to ASSH, 11 (79%) were published in peer-reviewed journals. Ten of the 14 accepted abstracts (71%) were considered positive, and there was no significant difference in publication rate between studies with positive ($n = 10$) and non-positive ($n = 4$) outcomes ($p = 1.000$).

Conclusions—The acceptance rate for negative outcomes studies regarding Kienbock's disease is higher than for other surgical disorders. This may indicate a relative decrease in positive outcome bias among published Kienbock's studies compared to other surgical disorders. However, the increasing positive outcome rate for published Kienbock's studies over time may suggest a trend of increasing publication bias among journals toward Kienbock's studies.

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Keywords

Kienbock's disease; systematic review; publication bias

Kienbock's disease, or avascular necrosis of the lunate, is classified as a “rare disease” by the Office of Rare Diseases (ORD) of the National Institutes of Health (NIH), and therefore affects less than 200,000 people in the U.S.[1] Since the initial description of this condition by Robert Kienbock in 1910, surgeons have described over 20 different procedures aimed at treating this disease.[2-4] The low prevalence and unclear etiology of Kienbock's disease present unique challenges with regards to conducting large, clinically meaningful comparative effectiveness trials. As a result, there is little evidence-based data to support any particular method of operative treatment, or to indicate their superiority over conservative measures.[3] Thus, hand surgeons may be inadvertently driven to treat Kienbock's patients with procedures that have the most recent or prevalent data presented in the literature, rather than based on objective comparative outcomes.

Publication bias refers to the tendency of researchers, peer reviewers, and journal editors to submit or accept manuscripts for publication based on the direction or strength of study findings.[5-6] This includes: 1) factors that influence the undertaking/performance of research (prepublication bias); 2) acceptance or rejection of an abstract/manuscript (publication bias), and; 3) acceptance or interpretation of previously published research as evidenced by citations in future research and inclusion in systematic reviews/meta-analyses (postpublication bias).[6-8] The most serious potential consequence of publication bias at any stage would be overestimation of treatment effects or risk-factor associations in published papers that could lead to inappropriate decisions concerning patient management or health policy.[9-11] Previous studies have demonstrated an association between positive outcomes, the presence of statistical significance, more advanced study design, study size, and increased likelihood of ultimate publication.[9-17] Due to the limited number of individuals affected with Kienbock's disease and the inherent difficulties associated with conducting research for rare disorders, the intense effort in hand surgery to treat this uncommon condition may be susceptible to publication bias.

Although several studies have assessed publication bias in various surgical specialties, there is a lack of specific data evaluating publication bias within the field of hand surgery.[9,13-16] The purpose of this study is to conduct a rigorous evaluation of all available literature regarding the surgical treatment of Kienbock's disease and identify potential areas of publication bias. Using data from published abstracts in both the English and non-English language, we will present a systematic review assessing study characteristics, quality of reporting, and direction of outcomes (positive or neutral/negative) for all published Kienbock's studies. In addition to our evaluation of published abstracts, we will collect the same data from abstracts presented at the 48th-60th annual scientific meeting of the American Society for Surgery of the Hand (ASSH; 1992-2004). By comparing the study characteristics and outcomes of unpublished abstracts to abstracts accepted for publication, we aim to determine factors that were positively or negatively associated with achieving ultimate publication.

Materials and Methods

Literature Search and Abstract Selection

Systematic searches of all available literature from the MEDLINE, EMBASE, and SCPOUS databases (1902-2009) were performed using the key words *Kienbock, avascular necrosis, revascularization, and lunate*. The resulting titles and abstracts were then screened to rule out non-primary, biomechanical, non-surgical, non-human, non-Kienbocks, anatomical, or diagnostic imaging studies. Studies evaluating pediatric patients or patients with concomitant joint, collagen, or vascular disease were also excluded. Relevant abstracts for analysis consisted of primary studies assessing outcomes for specific procedures used to treat Kienbock's disease. Abstracts containing multiple indications were included as long as the results for Kienbock's patients alone were able to be extracted. Due to the paucity of published research regarding Kienbock's disease, we included both English and non-English studies as long as the non-English studies contained a published English abstract.

In addition to our assessment of abstracts in published manuscripts, we also evaluated the same study characteristics and outcome parameters for abstracts accepted to the 48th to 60th annual scientific meeting of the ASSH (1992-2004). Abstracts prior to 1992 and abstracts from other hand conferences were not available for review. Publications corresponding to the accepted abstracts were identified through a computer search of the MEDLINE database using authors' names and key words from the title. We choose to review abstracts presented until the year 2004 based on previous research which demonstrated that over 90% of abstracts that will eventually lead to publication will publish within 5 years of abstract presentation.[12]

Data Extraction and Analysis

For each published manuscript and ASSH abstract included in our analysis, data regarding study characteristics (primary language, country of journal origin, study size), outcome direction (positive, negative, or neutral), and complications were collected. In addition, the following time-to-event information was collected for studies with available data: 1) length of patient enrollment, 2) mean follow-up time, and 3) time from completion of patient enrollment to publication. The presence of statistical analysis, designated by the presence of a p value (regardless of the value), was used as a crude indicator of study quality.

All manuscripts and abstracts were analyzed by two reviewers (L.S. and E.P.) with discrepancies resolved between each other by consensus. The results of each study were classified as positive, neutral, or negative. Positive outcomes were defined as present if the authors directly recommended a procedure or stated that the intervention was generally beneficial, irrespective of absolute outcome values or the presence of statistical significance; negative outcomes were defined as present when the authors advised against the intervention or presented only negative results; and neutral outcomes were defined by the absence of author recommendations or the presence of both positive and negative comments/ results.[13-16] For studies comparing more than one intervention for Kienbock's disease, outcomes were considered positive if the authors specifically recommended at least one of the procedures included in the study.

Statistical Analysis

Various categorical (primary language of study, country of study origin, procedure type, outcome, and presence of statistical analysis) and interval (number of Kienbock's wrists per study, complication rate, follow-up time, time to publication, and length of patient enrollment) data parameters were collected for each abstract. The statistical significance of relationships between categorical variables was evaluated using either chi-square tests (if all expected values were > 5) or Fischer's exact tests (if any expected value was ≤ 5). For comparison of mean interval data parameters, either a two-tailed t test (two groups) or analysis of variance (more than two groups) was performed. P values of less than 0.05 were considered statistically significant and all statistical analyses were performed using SAS statistical software (version 9.1; SAS, Cary, North Carolina).

Results

Published Kienbock's Abstracts

One thousand five hundred eighty seven citations were identified through the MEDLINE, EMBASE, and SCPOUS databases (Figure 1). Of the 253 relevant titles that underwent abstract review, 175 studies (124 English and 51 non-English) met all of our inclusion/exclusion criteria and formed the basis for our systematic review (Appendix A). Fourty-five articles were eliminated due to inextractable data for Kienbock's patients and thirty-three articles were eliminated because they lacked an associated English abstract. Although both English and non-English articles were primarily composed of studies from Europe (42% and 86%, respectively), English articles contained a greater proportion of studies from non-European countries ($p < 0.001$, Table 1). English articles were also more likely to include some form of statistical analysis (40% vs. 3%, $p < 0.001$, Table 1).

Overall, our data show that 53% of Kienbock's abstracts were associated with positive outcomes, which is much lower than the 74% positive outcome rate found among top orthopaedic and general surgery journals (Table 2).[13] Positive outcomes were associated with lower complication rates ($p = 0.006$, Table 3) and increased presence of statistical analysis ($p = 0.001$, Table 3). We found no correlation between positive outcomes and study of journal origin ($p = 0.071$), primary language of article ($p = 0.053$), study size ($p = 0.987$), and mean follow-up time ($p = 0.481$, Table 3). Of note, we also found no statistically significant association between negative outcomes and increased time to publication ($p = 0.747$) or length of patient enrollment ($p = 0.264$), which has been suggested in previous research regarding publication bias.[21]

Figure 2 demonstrates trends in surgical Kienbock's research over the past 40 years. As awareness of Kienbock's disease has grown, the number of primary clinical trials pertaining to Kienbock's disease has increased substantially from 11 studies during 1970-1979 to 71 studies during 2000-2009. Over time, results from published studies have become more positive (36% to 68%, $p = 0.007$) and are more likely to incorporate statistical analysis testing (0% to 55%, $p < 0.001$, Figure 2). These findings could be the result of improved management of Kienbock's disease and improved study quality over time. However, the

increasing positive outcome rate for published Kienbock's studies over time may suggest a trend of increasing publication bias among journals toward Kienbock's studies.

Abstracts Accepted to ASSH

Twenty-four abstracts pertaining to Kienbock's disease were identified from the 1992-2004 annual ASSH meetings (Table 4). Ten abstracts were considered inappropriate for review (2 diagnostic/imaging, 2 non-human, 1 non-operative, 5 inextractable data) and thus eliminated from our analysis. Of the 14 abstracts included in our study, 11 (79%) were ultimately published in peer-reviewed journals with an average of 2.2 years to publication. The presence of positive outcomes or statistical analysis had no significant impact on ultimate publication ($p = 1.000$, $p = 1.000$, respectively).

When comparing study characteristics of the 14 ASSH meeting abstracts with the manuscripts published during the same time period from our systematic review ($n = 128$), we found that abstracts presented at ASSH had a slightly higher positive outcome rate (71% vs 59%), a shorter average time to publication (2.2 years vs. 5.4 years), and were more likely to include some form of statistical analysis testing (50% vs 41%) compared to published manuscripts during the same time period. These results may indicate that Kienbock's studies presented at the ASSH have better outcomes and are of better quality than studies ultimately published in the literature which ultimately results in a faster time to publication.

Discussion

Ever since the advent of scientific and medical periodicals in the late 17th century, the publication of medical research has provided a basis for the common understanding of medical diseases and their associated treatment options. However, it was not until the early 1980's that direct evidence of publication bias within the medical literature became widely recognized.[5,9,25-31] Over the past three decades, medical researchers have become increasingly conscientious of the potential consequences of publication bias (overestimation of treatment effects and inappropriate risk factor associations) on patient management and health policy. Recent studies have demonstrated a clear bias toward publishing research with positive outcomes, increased sample size, and statistically significant results.[5,8,11,14,31] More specifically, evaluation of the surgical literature has found that top surgical journals are at increased risk for publication bias due to the low prevalence of high-level clinical studies and variable complication rates between different surgical centers.[13,23-24] This problem is further exacerbated for rare surgical disorders with poorly defined management strategies, such as Kienbock's disease. Due to the low prevalence, unknown etiology, poorly defined treatment algorithm, and lack of outcomes/complication data for Kienbock's disease, hand surgeons are more likely to base management decisions on randomly dispersed case reports/series in the published literature and presentations at national conferences. Thus, the presence of publication bias either in the published literature or among abstracts presented at national meetings has a greater propensity to directly affect patient care.

In this study, we present an analysis of all world literature (English articles and non-English articles with an English abstract) pertaining to the surgical management of Kienbock's disease. Overall, we did not find any evidence of positive outcome bias among published

Kienbock's manuscripts. This conclusion is supported by the low percentage of positive outcome studies for Kienbock's disease (53%) compared to other surgical disorders (74%), and the failure to demonstrate a relationship between positive outcomes in abstracts presented at annual ASSH meetings and achieving ultimate publication.[13] However, the lower positive outcome rate for Kienbock's studies could be the result of poorer expected outcomes for the surgical management of Kienbock's disease rather than decreased positive outcome bias among published Kienbock's abstracts.

Our results also demonstrated a higher publication rate of Kienbock's studies (79%) compared to the publication rate of other hand disorders (52%) presented at the ASSH annual meetings which could indicate potential selection bias at the national conference level prior to manuscript submission (pre-publication bias).[32] This theory is further supported by the slightly higher positive outcome rate (71% vs 59%), shorter average time to publication (2.2 years vs. 5.4 years), and increased likelihood of including some form of statistical analysis testing (50% vs 41%) among ASSH abstracts compared to published manuscripts during the same time period. However, the limited sample size of our analysis (14 ASSH abstracts) makes it impossible to draw any firm conclusions from these results and their interpretations should be reviewed with caution. Our goal of including this analysis in our study was to increase awareness among hand surgeons of potential selection bias among abstracts presented at national conferences and to encourage discretion when using these results to make patient management decisions.

Study Limitations

As with any systematic review, our study was limited by the quality of available literature. This limitation was especially challenging for Kienbock's disease. Forty-five published studies were unable to be incorporated into our analysis because they involved multiple procedures and/or multiple indications without providing specific outcomes data for Kienbock's patients. Of the remaining 175 published studies included in our analysis, most were small non-comparitive case series.

In an attempt to compensate for the lack of published Kienbock's studies and also to reduce publication bias within our own systematic review, we choose to include both English and non-English literature. However, because of language barriers, we were only able to review the English abstracts associated with non-English articles. This hindrance may have considerably reduced our sample size for various data parameters and consequently impaired our ability to determine statistically significant results.

Another limitation unique to studies analyzing positive outcome bias for specific conditions is the fact that various conditions will have different rates of positive outcomes based on inherent differences in management efficacy. For example, one would expect an inherently lower positive outcome rate for studies researching treatment for pancreatic cancer compared to studies researching management for distal radius fractures simply because distal radius fractures are easier to treat successfully. Thus, an apparent publication bias, as measured by positive outcome rate, does not necessarily imply a flaw in the editorial process.

For our study we were unable to determine whether the cause of our observed low positive outcome rate (53%) was the result of poorer expected outcomes among surgically managed Kienbock's patients or decreased publication bias among published Kienbock's studies. Other studies assessing publication bias for other conditions have attempted to correct for this issue by comparing study characteristics between published and non-published manuscripts or abstracts either accepted or not accepted to national conferences.[8,12] However, the authors of this study did not have access to non-published literature or ASSH abstract submissions prior to acceptance.

Due to the paucity of available literature, varied outcome measures, and our inability to review the entirety of non-English articles, we were unable to perform a comprehensive comparative analysis of surgical Kienbock's procedures. Although we recognize the need for a formal evidence-based comparative evaluation of Kienbock's procedures, the purpose of our analysis is to provide a systematic, critical review of the existing literature, identify any evidence of publication bias, and highlight research challenges facing rare surgical diseases.

Future Recommendations

Research regarding rare diseases has historically suffered from decreased attention, inadequate funding, and poor patient recruitment.[33-36] As a result, clinical studies of rare diseases often lack randomized controlled trials and formal statistical analyses, thus making them more prone to experience publication bias.[33-36] This means that physicians treating such rare diseases must form their clinical judgment solely on the basis of (potentially biased) observational studies, experience, and anecdote.[33] In 1993 the NIH founded the Office of Rare Diseases Research (ORDR), which coordinates and supports research of rare diseases.[1] Recent studies examining publication bias and challenges specific to rare disease research have promoted increased utility of the national clinical trial registry (www.clinicaltrials.gov), which provides information regarding all prospective clinical trials.[35-37] However, this design is poorly suited for many surgical disorders due to the predominance of retrospective outcome analysis. Furthermore, our study demonstrates that a considerable portion of surgical Kienbock's research is conducted outside the United States. We recommend the formation of an international patient database for Kienbock's disease that would provide standardized information regarding outcomes, complications, and follow-up for all surgical treatment methods. This would facilitate the development of clear evidence based outcomes regarding surgical management of this rare disease and enable determination of optimal treatment algorithms.

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Appendix A

English Abstracts

	Author	Year	Journal	Outcomes
1	Ozalp	2009	Archives of Orthopaedic and Trauma Surgery	Positive
2	Gay	2009	Plastic and Reconstructive Surgery	Positive
3	Mathoulin	2009	Microsurgery	Positive
4	Aly	2009	Orthopedics	Positive
5	Meena	2009	Indian Journal of Orthopaedics	Neutral
6	Croog	2008	Journal of Hand Surgery (American)	Positive
7	Waitayawinyu	2008	Journal of Hand Surgery (American)	Positive
8	Altay	2008	International Orthopaedics	Positive
9	Arora	2008	Journal of Hand Surgery (American)	Positive
10	Streich	2008	International Orthopaedics	Positive
11	Watanabe	2008	Journal of Bone and Joint Surgery (American)	Positive
12	Lumsden	2008	Journal of Hand Surgery (American)	Positive
13	Jones	2008	Journal of Hand Surgery (Euroepan Volume)	Neutral
14	Kawoosa	2007	International Orthopaedics	Neutral
15	Hermans	2007	Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery	Neutral
16	Tatebe	2007	Journal of Hand Surgery (American)	Positive
17	Raven	2007	Clinical Orthopaedics and Related Research	Positive
18	Tambe	2007	Acta Orthopaedica Belgica	Negative
19	El-Mowafi	2006	Acta Orthopaedica Belgica	Neutral
20	Schweizer	2006	Journal of Hand Surgery (American)	Neutral
21	Tatebe	2006	Hand Surgery	Positive
22	Gong	2006	Journal of Bone and Joint Surgery (British)	Positive
23	Vanden Dungen	2006	Chirurgie de la Main	Negative
24	Lu	2006	Annals of Plastic Surgery	Positive
25	Moran	2005	Journal of Hand Surgery (American)	Positive
26	Yasuda	2005	Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery	Positive
27	Daecke	2005	Journal of Hand Surgery (American)	Neutral
28	Daecke	2005	Journal of Hand Surgery (American)	Positive
29	Zafra	2005	Acta Orthopaedica Belgica	Positive
30	Tambe	2005	International Orthopaedics	Positive
31	Zenzai	2005	Journal of Hand Surgery (British and European Volume)	Positive
32	DeSmet	2005	Journal of Hand Surgery (British and European Volume)	Positive
33	Sakai	2004	Hand Surgery	Neutral
34	Meier	2004	Journal of Hand Surgery (British and European Volume)	Positive
35	Yajima	2004	Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery	Positive

	Author	Year	Journal	Outcomes
36	Thomas	2004	Journal of Hand Surgery (American)	Positive
37	Leblebicioglu	2003	Arthroscopy	Neutral
38	Watson	2003	Journal of Hand Surgery (American)	Neutral
39	Koh	2003	Journal of Hand Surgery (American)	Positive
40	Kuhlman	2003	Acta Orthopaedica Belgica	Positive
41	Gabl	2003	European Surgery	Positive
42	Chillemi	2003	Journal of Orthopaedics and Traumatology	Positive
43	Iwasaki	2002	Journal of Bone and Joint Surgery (British)	Positive
44	Wada	2002	Journal of Hand Surgery (British and European Volume)	Neutral
45	Laing	2002	European Journal of Orthopaedic Surgery and Traumatology	Positive
46	Gabl	2002	Journal of Hand Surgery (British and European Volume)	Neutral
47	Oishi	2002	Plastic and Reconstructive Surgery	Positive
48	Minami	2002	Current Opinion in Orthopaedics	Positive
49	Soejima	2002	Journal of Hand Surgery (American)	Positive
50	Wintman	2001	Orthopedics	Positive
51	Takase	2001	Journal of Bone and Joint Surgery (American)	Positive
52	Illarramendi	2001	Journal of Hand Surgery (American)	Positive
53	Kakinoki	2001	Hand Surgery	Neutral
54	Bengoechea-Beeby	2001	Journal of Hand Surgery (American)	Neutral
55	Sauerbier	2000	Annals of Plastic Surgery	Positive
56	Lamas	2000	Journal of Hand Surgery (American)	Neutral
57	Salmon	2000	Journal of Bone and Joint Surgery (British)	Positive
58	Makino	2000	Journal of Reconstructive Microsurgery	Neutral
59	Menth-Chiari	1999	Arthroscopy	Positive
60	Ueba	1999	Journal of Orthopaedic Science	Neutral
61	Nakamura	1998	Journal of Hand Surgery (British and European Volume)	Positive
62	Kaarela	1998	Journal of Hand Surgery (British and European Volume)	Negative
63	Delaere	1998	Journal of Hand Surgery (British and European Volume)	Negative
64	Moneim	1998	Iowa Orthopaedic Journal	Positive
65	Yajima	1998	Journal of Hand Surgery (American)	Positive
66	Garcia-Elias	1998	Annales de Chirurgie de la Main	Neutral
67	Shayfer	1998	Orthopedics	Neutral
68	Steenwerckx	1997	Acta Orthopaedica Belgica	Neutral
69	Carroll	1997	Clinical Orthopaedics and Related Research	Neutral
70	Quenzer	1997	Journal of Hand Surgery (American)	Neutral
71	Trail	1996	Journal of Hand Surgery (British and European Volume)	Positive
72	Watson	1996	Journal of Hand Surgery (American)	Positive
73	Zelouf	1996	Journal of Hand Surgery (American)	Positive
74	Guo	1996	Annals of Plastic Surgery	Neutral

	Author	Year	Journal	Outcomes
75	Miura	1996	Journal of Hand Surgery (American)	Positve
76	Wheatley	1996	Annals of Plastic Surgery	Positive
77	Rhee	1996	Journal of Korean Medical Science	Neutral
78	Sennwald	1995	Journal of Hand Surgery (American)	Positive
79	DeSmet	1995	Acta Orthopaedica Belgica	Negative
80	Tomaino	1994	Journal of Hand Surgery (American)	Neutral
81	Bochud	1994	Journal of Hand Surgery (British and European Volume)	Neutral
82	Yajima	1994	Journal of the Japanese Orthopaedic Association	Neutral
83	Begley	1994	Journal of Hand Surgery (American)	Positive
84	Minami	1994	Journal of Hand Surgery (American)	Positive
85	Amillo	1993	International Orthopaedics	Positive
86	Inoue	1992	Archives of Orthopaedic and Trauma Surgery	Neutral
87	Voche	1992	Journal of Hand Surgery (British and European Volume)	Positive
88	O'Flanagan	1992	Journal of the Royal College of Surgeons of Edinborough	Negative
89	Inoue	1992	Acta Orthopaedica Scandinavica	Neutral
90	Nakamura	1991	Journal of Bone and Joint Surgery (American)	Positive
91	Weiss	1991	Journal of Bone and Joint Surgery (American)	Positive
92	Rock	1991	Journal of Hand Surgery (American)	Positive
93	Inoue	1990	Acta Orthopaedica Scandinavica	Positive
94	Hasselgren	1990	Journal of Hand Surgery (British and European Volume)	Positive
95	Alexander	1990	Journal of Hand Surgery (American)	Negative
96	Kawai	1988	Journal of Bone and Joint Surgery (American)	Neutral
97	Viljakka	1987	Acta Orthopaedica Scandinavica	Neutral
98	Schattenkerk	1987	Acta Orthopaedica Scandinavica	Neutral
99	Ekerot	1986	Scandinavian Journal of Plastic and Reconstructive Surgery	Neutral
100	Kato	1986	Journal of Hand Surgery (American)	Positive
101	Evans	1986	Journal of Hand Surgery (British and European Volume)	Neutral
102	Watson	1985	Journal of Hand Surgery (American)	Neutral
103	Blanco	1985	Journal of Hand Surgery (American)	Neutral
104	Backaert	1985	Acta Orthopaedica Belgica	Neutral
105	Sundberg	1984	Clinical Orthopaedics and Related Research	Neutral
106	Pardini	1984	International Orthopaedics	Neutral
107	Ishiguro	1984	Journal of the Japanese Orthopaedic Association	Positive
108	Eiken	1984	Scandinavian Journal of Plastic and Reconstructive Surgery	Positive
109	Kinnard	1983	Canadian Journal of Surgery	Positive
110	Armistead	1982	Journal of Bone and Joint Surgery (American)	Positive
111	Ramakrishna	1982	Journal of Bone and Joint Surgery (American)	Neutral
112	Almquist	1982	Journal of Hand Surgery (American)	Neutral
113	Lichtman	1982	Journal of Hand Surgery (American)	Positive

	Author	Year	Journal	Outcomes
114	Hedeboe	1982	Scandinavian Journal of Plastic and Reconstructive Surgery	Positive
115	Bertini	1982	Italian Journal of Orthopaedics and Traumatology	Positive
116	Stark	1981	Journal of Bone and Joint Surgery (American)	Negative
117	Grassi	1978	Italian Journal of Orthopaedics and Traumatology	Positive
118	Inglis	1977	Journal of Bone and Joint Surgery (American)	Positive
119	Lichtman	1977	Journal of Bone and Joint Surgery (American)	Neutral
120	Roca	1976	Journal of Bone and Joint Surgery (American)	Neutral
121	Barber	1974	Journal of Bone and Joint Surgery (British)	Positive
122	Codega	1973	International Surgery	Neutral
123	Nahigian	1970	Journal of Bone and Joint Surgery (American)	Neutral
124	Graner	1966	Journal of Bone and Joint Surgery (American)	Positive

Non-English Abstracts

	Author	Year	Journal	Outcomes
1	Von-Maydell	2008	Handchirurgie, Mikrochirurgie, Plastische Chirurgie	Neutral
2	Lecomte	2007	Revue de Chirurgie Orthopedique et Reparatrice de l'Appareil Moteur	Neutral
3	Baronetti	2006	Minerva Ortopedica e Traumatologica	Neutral
4	Khorbi	2005	Tunisie Medicale	Neutral
5	Adel	2005	Tunisie Medicale	Positive
6	Amillo-Garayoa	2005	Revista de Ortopedia y Traumatologia	Positive
7	Lu	2003	Chinese Medical Journal	Positive
8	Welby	2003	Chirurgie de la Main	Negative
9	Das Gupta	2003	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
10	Altay	2002	Artroplasti Artroskopik Cerrahi	Positive
11	Sauerbier	2001	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
12	Trankle	2000	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
13	Siala	2000	Revue de Chirurgie Orthopedique et Reparatrice de l'Appareil Moteur	Positive
14	Jiang	1999	Chinese Journal of Reparative and Reconstructive Surgery	Positive
15	Dautel	1999	Main	Neutral
16	Sauerbier	1998	Langenbecks Archiv für Chirurgie. Supplement. Kongressband. Deutsche Gesellschaft für Chirurgie. Kongress	Positive
17	Bartelmann	1998	Handchirurgie Mikrochirurgie Plastische Chirurgie	Negative
18	Schulz	1998	Handchirurgie Mikrochirurgie Plastische Chirurgie	Neutral
19	Renner	1998	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
20	Wustner-Hofmann	1997	Handchirurgie Mikrochirurgie Plastische Chirurgie	Neutral
21	Staudenmaier	1997	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
22	Thomas	1997	Annales de Chirurgie de la Main et du Membre Superieur	Neutral

	Author	Year	Journal	Outcomes
23	Ducarmois	1997	Annales de chirurgie de la main et du membre supérieur	Neutral
24	Garbuio	1996	Annales de chirurgie de la main et du membre supérieur	Neutral
25	Wachtl	1994	Schweizerische Rundschau fur Medizin Praxis	Negative
26	Gomis	1994	Revue de Chirurgie Orthopedique et Reparatrice de l'Appareil Moteur	Positive
27	Allieu	1991	Annales de Chirurgie de la Main et du Membre Superieur	Positive
28	Allieu	1991	Annales de chirurgie de la main et du membre supérieur	Positive
29	Voche	1991	Revue de Chirurgie Orthopedique et Reparatrice de l'Appareil Moteur	Positive
30	Ham	1990	Nederlands Tijdschrift voor Geneeskunde	Neutral
31	Buck-Gramcko	1990	Handchirurgie Mikrochirurgie Plastische Chirurgie	Negative
32	Kawai	1990	Annales de Chirurgie de la Main et du Membre Superieur	Neutral
33	Shibata	1989	Journal of the Japanese Orthopaedic Association	Neutral
34	Lesur	1989	Revue de chirurgie orthopédique et réparatrice de l'appareil moteur	Neutral
35	Yang	1989	Chinese Journal of Surgery	Neutral
36	Ehall	1989	Handchirurgie Mikrochirurgie Plastische Chirurgie	Positive
37	Prommersberger	1988	Handchirurgie Mikrochirurgie Plastische Chirurgie	Neutral
38	Kern	1988	Zeitschrift fur Orthopadie und Ihre Grenzgebiete	Neutral
39	Bruser	1986	Handchirurgie Mikrochirurgie Plastische Chirurgie	Neutral
40	Rajani	1985	Ugeskrift for Laeger	Positive
41	Erbs	1984	Handchirurgie Mikrochirurgie Plastische Chirurgie	Neutral
42	Razemon	1984	Chirurgie	Positive
43	Schmitt	1984	Zeitschrift fur Orthopadie und Ihre Grenzgebiete	Positive
44	Saffar	1982	Annales de Chirurgie de la Main	Neutral
45	Roullet	1982	Annales de Chirurgie de la Main	Negative
46	Comtet	1982	Annales de chirurgie de la main	Neutral
47	Naett	1981	Handchirurgie	Neutral
48	Kerschbaumer	1981	Orthopade	Positive
49	Kerschbaumer	1979	Handchirurgie	Negative
50	Codega	1973	Polski Przeglad Chirurgiczny	Neutral
51	Sommelet	1970	Revue de chirurgie orthopédique et réparatrice de l'appareil moteur	Neutral

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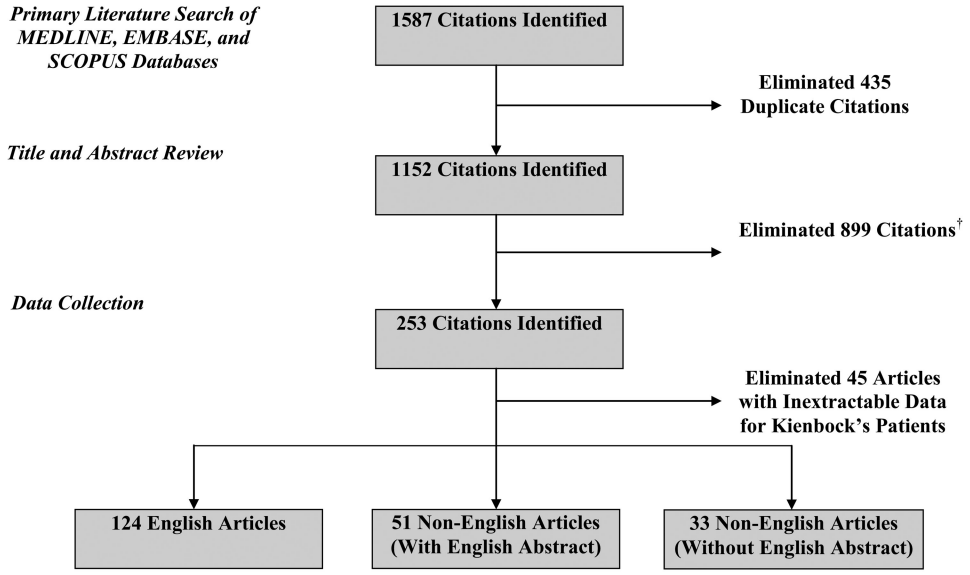

Figure 1. Flowchart of primary database search and data collection

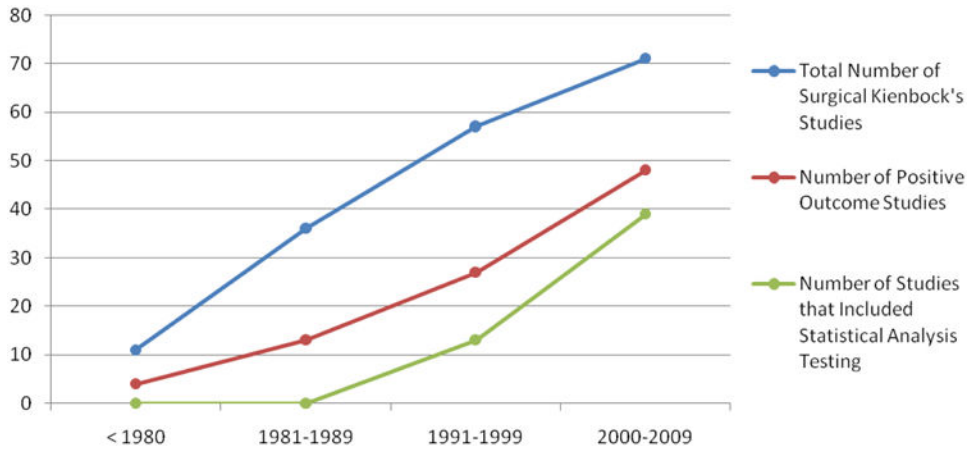

Figure 2. Trends in Surgical Kienbock's Research Over Time

Table 1
Study Characteristics of Published Kienbock's Abstracts

	Total Abstracts (n = 175)	English Article (n = 124)	Non-English Article (n = 51)	P Value
Country of Journal Origin				
US	63	63 (51%)†	0 (0%)‡	<0.001δ
Europe	96	52 (42%)†	44 (86%)‡	
Asia	12	8 (6%)†	4 (8%)‡	
Other	4	1 (1%)†	3 (6%)‡	
Procedure Type*				
Joint Leveling	51	32 (25%)†	19 (33%)‡	0.257ζ
Implant/Transposition Arthroplasty	36	26 (20%)†	10 (17%)‡	
Partial Wrist Fusion	25	17 (13%)†	8 (14%)‡	
Vascular Bone Graft/Pedicle	18	10 (8%)†	8 (14%)‡	
Salvage	22	18 (14%)†	4 (7%)‡	
Other	36	27 (21%)†	9 (16%)‡	
Overall Outcomes				
Positive	92	71 (57%)†	21 (41%)‡	0.053ζ
Neutral/Negative	83	53 (43%)†	30 (59%)‡	
Mean Number of Wrists per Study$^{\alpha}$	19.8	18.8	22.3	0.415ε
Complication Rate$^{\beta}$	17.7%	17.0%	22.7%	0.435ε
Mean Follow-Up (months) $^{\gamma}$	71	68	83	0.276ε
Presence of Statistical Analysis	52	50 (40%)†	2 (4%)‡	<0.001ζ
Time to Publication (years)$^{\pi}$	5.1	5.4	3.5	0.103ε
Length of Patient Enrollment (years) $^{\Omega}$	10.0	10.4	7.3	0.117ε

†Data presented as number of English abstracts (% of total English abstracts)

‡Data presented as number of non-English abstract (% of total non-English abstracts)

*For studies including more than one surgical procedure, separate entries were made for each procedure type

$^{\alpha}$Only includes wrists corresponding to surgical Kienbock's patients

$^{\beta}$Data presented as the average complication rate for studies with available data; Reported in 78 of 124 English articles (63%) and 11 of 51 non-English articles (22%)

$^{\gamma}$Data reported in 110 of 124 English articles (89%) and 30 of 51 non-English articles (59%)

$^{\pi}$Data presented as time from end of patient enrollment to publication; Reported in 75 of 124 English articles (60%) and 12 of 51 non-English articles (24%)

$^{\Omega}$Data reported in 75 of 124 English articles (60%) and 12 of 51non-English articles (24%)

[δ] Statistical analysis using Fisher's exact test

[ε] Statistical analysis using two-tailed t-test

[ζ] Statistical analysis using chi-square test

Table 2
Factors Associated with Positive Outcomes

	Total Abstracts (n = 175)	Positive Outcomes (n = 92)	Non-Positive Outcomes (n = 83)	P value
Country of Journal Origin				
US	63	40 (43%)†	23 (28%)‡	0.071ε
Europe	96	45 (49%)†	51 (61%)‡	
Asia	12	4 (4%)†	8 (10%)‡	
Other	4	3 (3%)†	1 (1%)‡	
Language of Article				
English Article	124	71 (77%)†	53 (64%)‡	0.053ε
Non-English Article with English Abstract	51	21 (23%)†	30 (36%)‡	
Mean Number of Wrists per Study α	19.8	19.8	19.9	0.987δ
Complication Rate β	17.7%	12.4%	25.8%	0.006δ
Mean Follow up Time γ	71.2	67.7	75.5	0.481δ
Presence of Statistical Analysis	52	37 (40%)	15 (18%)	0.001ε
Time to Publication (years) π	5.1	5.2	5.0	0.747δ
Length of Patient Enrollment (years)Ω	10.0	10.8	9.0	0.264δ

†Data presented as number of positive abstracts (% of total positive abstracts)

‡Data presented as number of negative abstract (% of total negative abstracts)

αOnly includes wrists corresponding to surgical Kienbock's patients

βData presented as the average complication rate for studies with available data; Reported in 54 of 92 studies with positive outcomes (59%) and 35 of 83 studies with non-positive outcomes (42%)

γData reported in 78 of 92 studies with positive outcomes (85%) and 62 of 83 studies with non-positive outcomes (75%)

πData presented as time from end of patient enrollment to publication; Reported in 47 of 92 studies with positive outcomes (51%) and 40 of 83 studies with non-positive outcomes (48%)

ΩData reported in 47 of 92 studies with positive outcomes (51%) and 40 of 83 studies with non-positive outcomes (48%)

δStatistical analysis using two tailed t-test

εStatistical analysis using chi-squared test

Table 3

Outcomes, Complications, and Quality of Reporting According to Country of Journal Origin

	US (n = 63)	Europe (n = 96)	Asia (n = 12)	Other (n = 4)	P value
Overall Outcomes					
Positive[†]	40 (63%)	45 (47%)	4 (33%)	3 (75%)	0.071[δ]
Neutral/Negative[†]	23 (37%)	51 (53%)	8 (67%)	1 (25%)	
Mean Number of Wrists per Study[‡]	20.7	19.4	20.4	14.8	0.939[ε]
Complication Rate [α]	12.3%	24.3%	7.6%	0.1%	0.067[ε]
Mean Follow-Up [β]	73	72	55	66	0.894[ε]
Statistical Analysis Present[†]	29 (46%)	20 (21%)	3 (25%)	0 (0%)	0.004[δ]
Time to Publication (years) [γ]	5.8	4.5	5.4	NA	0.267[ε]
Length of Patient Enrollment (years) [π]	11.6	8.1	12.3	NA	0.093[ε]

[†]Data reported as number of abstracts (% of total abstracts for given geographic region)

[‡]Only includes wrists corresponding to Kienbock's patients

[α]Data presented as the average complication rate for studies with available data; Reported in 42 of 63 US studies (67%), 42 of 96 European studies (44%), 3 of 12 Asian studies (25%), and 2 of 4 studies published in another country (50%)

[β]Data reported in 59 of 63 US studies (94%), 68 of 96 European studies (71%), 9 of 12 Asian studies (75%), and 4 of 4 studies published in another country (100%)

[γ]Data presented as time from end of patient enrollment to publication; Reported in 38 of 63 US studies (60%), 42 of 96 European studies (44%), 7 of 12 Asian studies (58%), and 0 of 4 studies published in another country (0%)

[π]Data reported in 38 of 63 US studies (60%), 42 of 96 European studies (44%), 7 of 12 Asian studies (58%), and 0 of 4 studies published in another country (0%)

[δ]Statistical analysis using Fisher's exact test

[ε]Statistical analysis using ANOVA

Table 4

Study Characteristics of Kienbock's Abstracts Presented at the 1992-2004 ASSH Annual Meeting†

Variables	Number of Abstracts	Published (%)	P value
Country of Study Origin			
US	6	5 (83%)	1.000‡
Other	8	6 (75%)	
Total Number of Wrists			
< 20	9	8 (89%)	0.506‡
> 20	5	3 (60%)	
Result			
Positive	10	8 (80%)	1.000‡
Neutral/Negative	4	3 (75%)	
Statistical Analysis			
No	7	5 (71%)	1.000‡
Yes	7	6 (86%)	
Complications			
Not recorded	7	7 (100%)	0.096‡
≤ 10%	3	2 (67%)	
> 10%	4	2 (50%)	

† 14 abstracts were included in our analysis; 10 abstracts were eliminated (2 diagnostic/imaging, 2 non-human, 1 non-operative, 5 inextractable data for Kienbock's patients or for specific procedure)

‡ Statistical analysis using Fisher's exact test