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Intracranial Imaging of Uncommon Diseases Is More Frequently Reported in Clinical Publications Than in Radiology Publications

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ABSTRACT

BACKGROUND AND PURPOSE: Descriptions of uncommon diseases with intracranial imaging abnormalities are often difficult to find in the radiology literature. We hypothesized that reported imaging findings of such conditions in the recent literature were more frequent in clinical compared with radiology journals.

MATERIALS AND METHODS: PubMed searches from December 1, 2007 to December 1, 2012 were performed for 5 uncommon CNS diseases with intracranial imaging manifestations: 1) Susac syndrome; 2) amyloid β–related angiitis; 3) Parry-Romberg syndrome/en coup de sabre; 4) transient lesion of the splenium of the corpus callosum; and 5) reversible cerebral vasoconstriction syndrome. Articles were classified as a case report, case series, or original research. Journals were categorized as radiology or clinical. The 1- and 5-year Impact Factors of the journals were recorded.

RESULTS: Two hundred two articles were identified for the 5 diseases, including 151 (74%) case reports, 26 case series (13%), and 25 original research articles (13%); 179 (89%) were published in nonradiology journals, compared with 23 (11%) in radiology journals. There was no significant difference between the mean 1- and 5-year Impact Factors of the radiology and clinical journals.

CONCLUSIONS: Recent reports of the selected uncommon diseases with intracranial manifestations are more frequent in clinical journals when compared with dedicated radiology publications. Most publications are case reports. Radiologists should review both radiology and clinical journals when reviewing imaging features of uncommon diseases affecting the brain. Lack of reporting on such disease in the radiology literature may have significant practice, educational, and research implications for the radiology community.

ABBREVIATIONS: ABRA = amyloid β–related angiitis; RCVS = reversible cerebral vasoconstriction syndrome

The frequency with which the radiology versus the clinical literature reports on the imaging findings and techniques of uncommon CNS diseases is not known. Although little has been published about the rates at which radiologists read specific medical journals, there is some evidence that radiologists tend to read radiology journals and that specialists tend to focus on clinical journals within their respective areas.[1-3] There is also evidence that journal articles are a preferred source of information for clinical decision-making by physicians.[2] It is important to characterize the avenues of knowledge dissemination, to direct radiologists and clinicians to the most likely primary resources that can help optimize management of patients with an uncommon CNS disease.

In our day-to-day clinical practice, we have anecdotally found much of the relevant information on imaging findings about several uncommon CNS diseases in the clinical rather than the radiologic literature. However, this perception has not been confirmed with a systematic study of the medical literature. Additionally, the type of articles in which imaging of uncommon CNS disease is reported is not known but could be useful to investigate to increase understanding of the source of reported imaging findings. Many of the purported imaging features of uncommon CNS diseases may be found within case reports rather than within original research, in part because of low prevalence of disease and inherent difficulty with patient accrual. However, case reports have faced high rejection rates within the radiology literature, and some radiology journals no longer consider case reports for publication in recent years,[4] even though a survey has indicated that many radiologists find case reports useful.[5]

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To further investigate these unresolved questions, we selected 5 uncommon diseases with potentially important intracranial imaging manifestations. We hypothesized that reported imaging findings of such conditions in the recent literature were more frequent in clinical compared with radiology journals.

MATERIALS AND METHODS

CNS Conditions

Five uncommon conditions with important intracranial imaging manifestations were selected for review: Susac syndrome, transient lesion of the splenium of the corpus callosum, amyloid β–related angiitis (ABRA), Parry-Romberg syndrome/en coup de sabre, and reversible cerebral vasoconstriction syndrome (RCVS). Although the precise incidence of these conditions is not known with certainty, they were selected because 1) imaging plays an important role in diagnosis and/or management and 2) these are common enough that the authors have cumulatively seen cases of each in our subspecialized academic practice but evaluate each one infrequently. Multiple unrelated conditions were selected to evaluate for overall patterns in publication of topics relevant to neuroradiology rather than the publication pattern of a single condition. Because Parry-Romberg syndrome and en coup de sabre are now considered to be related along the same spectrum of morphea by many authorities in dermatology,[6] they were considered together as Parry-Romberg syndrome/en coup de sabre.

PubMed Search

PubMed searches of the English literature from December 1, 2007 to December 1, 2012 were performed for the aforementioned conditions. We evaluated topics over a 5-year period to capture an adequate number of publications for analysis but limited our search to these recent years to ensure results are representative of current patterns of publication. All searches were performed between December 1, 2012 and December 10, 2012. The condition names and common synonyms were used as search terms to capture most of the relevant literature. The following search terms were used with an English language–only modifier: 1) Susac syndrome: "Susac syndrome OR Susac's syndrome," 2) transient lesion of the splenium of the corpus callosum: "(transient lesion OR reversible lesion) AND corpus callosum," 3) ABRA: "amyloid beta-related angiitis OR ABRA OR amyloid vasculitis," 4) Parry-Romberg syndrome/en coup de sabre: "Parry-Romberg syndrome OR en coup de sabre," and 5) RCVS: "reversible cerebral vasoconstriction syndrome OR RCVS OR Call-Fleming."

Inclusion Criteria

Included case reports or case series contained, at minimum, a description of the imaging findings as well as at least 1 figure demonstrating a radiologic image of the brain. A report of multiple cases with common diagnosis and pure description of findings was categorized as a case series. A publication with multiple cases that included a clearly defined systematic method of analysis of an imaging finding/technique or other pertinent aspect of the disease was designated original research. Included original research contained, at minimum, description of imaging findings of some included patients, though an actual figure of an example of an imaging finding was not required. Included intracranial imaging was cross-sectional (either CT or MR imaging), transcranial Doppler, and/or angiographic (conventional DSA, MRA, or CTA). Original research articles that studied patients both with and without the target diagnosis, such as those with comparison of patients with ABRA and primary CNS vasculitis, were included. Imaging of the face without intracranial imaging, such as reports of extracranial head and neck findings in Parry-Romberg syndrome/en coup de sabre alone, was not sufficient for inclusion because the focus of this study was on intracranial findings.

Exclusion criteria were 1) wrong topic captured by search terms; 2) animal study; 3) review article, commentary, or editorial without concomitant incremental case report; 4) no images of the brain included for case reports or case series; 5) no description of imaging findings for an original research article; and 5) full-text not available when insufficient information for analysis was available in the abstract. The full text was analyzed whenever possible. For articles in which there was no abstract or insufficient information available within the abstract, an attempt was made to obtain the full text by our institutional librarians from the publisher or interlibrary loan. In these cases, if the librarians were unable to obtain the full text from the publisher without charge, the article could not be analyzed and was excluded.

Literature Search Analysis and Author Experience

The journal for each included article was recorded and categorized as radiology or clinical with consensus agreement among the authors. Radiology journals were those with a clear primary focus of reporting imaging findings/techniques. The authors reviewed all journals and arrived at consensus designation of each journal as in either the radiology or clinical category. A description of the type of imaging and imaging analysis in each original article was recorded. The articles within the most common radiology journal were examined to determine if they would fulfill current publication criteria (as of April 1, 2013). Additionally, although review, commentary, and editorial articles were not grouped with original reports and studies for the formal analysis, these could also serve as a source of information for radiologists and clinicians. Therefore, the journals of these publications were also categorized as radiology or clinical.

Six of the authors (V.T.L., D.F.B., C.H.H., F.E.D., K.M.S., L.J.E.) are board-certified radiologists with 1, 1, 2, 4, 5, and 7 years of postfellowship radiology experience, respectively. Four of the authors (C.H.H., F.E.D., K.M.S., L.J.E. have obtained a Certificate of Added Qualification in neuroradiology. One author (D.A.D.) is a radiology resident. Two of the authors (D.F.B. and C.H.H.) also have board certification in clinical neurology, with 5 years of clinical neurology experience each.

The affiliation of each author of the included articles was noted. Articles were categorized as having either 1) at least 1 radiologist as an author or 2) no radiologist listed as an author. If a radiologist author was not identified but the affiliation of every author could not be confirmed, the article was excluded from the author analysis. If at least 1 radiologist as an author was identified, it was included regardless of the availability or type of other author affiliations.

The 2011 1-year and 5-year Impact Factors were recorded for each journal as a measure of frequency of journal citation. The Impact Factors were obtained from an on-line data base, *Journal Citation Reports,* which is published by Thomson Reuters.[7] Journals that had no recorded Impact Factors were assigned Impact

Factor values of 0 for the purpose of data analysis. Mean Impact Factors of the radiology and clinical journals for each included article were compared with a paired *t* test. The mean Impact Factors were weighted to account for some journals having more than 1 article identified within this literature analysis; 95% CIs were calculated. $P < .05$ was considered statistically significant.

RESULTS

Literature Search

Four-hundred seventy-one publications were captured by the search terms. Two hundred sixty-nine were excluded from analysis for the following reasons: 1) wrong topic captured by search terms ($n = 160$, 59%); 2) animal study ($n = 3$, 1%); 3) review or commentary without incremental case report ($n = 51$, 19%); 4) no CNS images provided for a case report or series ($n = 45$, 17%); 5) full text unavailable when insufficient information was available in the abstract ($n = 10$, 4%); and 6) no imaging description for original research articles ($n = 0$, 0%).

Two hundred two publications (43% of 471) were identified within 12 radiology and 80 clinical journals. The most common radiology and clinical journals are listed in Table 1. The most common radiology journal was the *American Journal of Neuroradiology*, whereas the most common clinical journal was the *Journal of Neurologic Sciences*. Three of 5 (60%) of the articles in the *American Journal of Neuroradiology* were case reports or series with <5 patients and would no longer be considered for publication in this journal after July 1, 2011, whereas 2 (40%) were case series with >5 patients and would still be considered.[8] A complete list of the journals identified and designation as radiology or clinical is catalogued in the On-line Appendix.

Table 2 details the categorization of publication of the clinical and radiology articles. The final study sample of 202 publications included 151 (74%) case reports, 26 case series (13%), and 25 original research articles (13%). Overall, 179 (89%) were published in nonradiology journals compared with 23 (11%) in radiology journals.

Table 1: The 3 most common radiology and clinical journals with the included diagnoses

Journal	No. of Articles
Radiology	
1) *AJNR Am J Neuroradiol*	5
2) *J Neuroimaging*	4
3) *Interv Neuroradiol*	3
Clinical	
1) *J Neurol Sci*	16
2) *Cephalgia*	13
3) *Neurol Sci*	9

Table 2: Frequency of identified publication in radiology and clinical journals

Journal Type	PRS	ABRA	TLCC	Susac	RCVS	Overall
Radiology						
Case reports	4	0	6	3	5	18
Case series	0	0	4	0	1	5
Original research	0	0	0	0	0	0
All publication categories	4	0	10	3	6	23
Clinical						
Case reports	17	8	35	21	52	133
Case series	3	1	7	2	8	21
Original research	1	4	1	4	15	25
All publication categories	21	13	43	27	75	179

Note:—PRS indicates Parry-Romberg syndrome; TLCC, transient lesion of the splenium of the corpus callosum.

Case reports were the most common form of publication for articles in both the radiology and the clinical literature, accounting for 18 of 23 (78%) and 133 of 179 (75%) of radiology and clinical articles, respectively. Additionally, 47 of 51 (92%) separate review, commentary, or editorial articles were within clinical journals.

A brief description of the identified original research articles is listed in the On-line Table. All 25 (100%) original research articles were in the clinical literature. Fifteen (60%) of the original research articles addressed RCVS, though each of the 5 diagnoses had at least 1 original research article. The role of imaging within these articles varied and included articles with a primary aim of characterizing imaging findings such as serial MRA examinations with RCVS[9] or MR imaging findings of Susac syndrome at 7T,[10] as well as articles primarily assessing an imaging technique or sequence, such as diffusion tensor imaging in Susac syndrome.[11]

Author Affiliations

In 19 of 202 (9%) of articles, all in clinical journals, a radiologist author could not be confirmed and complete lists of author affiliations could not be found. Complete lists of author affiliations were available for 183 of 202 (91%) articles. Radiologists were authors in 98 of these 183 (54%) articles, including 22 of 23 (96%) articles in radiology journals and 76 of 160 (48%) articles in clinical journals.

Impact Factors

There was no significant difference between the mean 1- and 5-year Impact Factors (*P* values of .29 and .39, respectively) between radiology and clinical journals (Table 3). One-year Impact Factors did not exist for the journals of 1 of 23 (4%) articles in the radiology literature and for the journals of 21 of 179 (12%) articles in the clinical literature. Five-year Impact Factors did not exist for 2 of 23 (9%) articles in radiology journals and 31 of 179 (17%) articles in clinical journals.

DISCUSSION

In the present study, identified reports and original research of imaging features of each of the selected uncommon conditions with brain pathology were more frequent in the clinical compared with the radiology literature. Radiologists are under-represented as authors on such publications in the clinical literature. These findings have numerous implications for neuroradiology practice and patient care.

Description and analysis of imaging findings of CNS disease in the clinical literature is important for several reasons. Clinicians should know the utility of various neuroradiologic techniques to ensure the most appropriate test is requested for a given clinical scenario and should learn about new techniques as they become available. With many CNS pathologies, imaging findings can help direct treatment, can be of prognostic significance, and can help to assess response to clinical treatment.

Clinicians aware of reports of imaging findings of neurologic disease in the clinical literature will probably expect consulted neuroradiologists to have knowledge of these imaging findings. If the primary targeted audience of the journals containing these reports is highly skewed toward physicians other than radiologists, it is possible that these imaging findings, or

Table 3: Mean Impact Factors of included radiology and clinical journals

	1-Year Impact Factor		5-Year Impact Factor	
Journal Type	Mean (95% CI)	*P* Value	Mean (95% CI)	*P* Value
Radiology	1.80 (1.25–2.36)	.29	1.87 (1.20–2.54)	.39
Clinical	2.84 (2.17–3.51)		2.66 (2.02–3.29)	

Note:—Journals with no listed impact factor were assigned values of "0" for the analysis in this table.

even the existence of the condition, will be under-recognized by radiologists. In fact, under-recognition of Susac syndrome by radiologists has been cited by Dr Susac.[12] Radiologists are potentially the first physicians to encounter findings suggestive of a rare CNS condition and have great potential to affect management. Specifically, radiologist under-recognition could initiate an incorrect, possibly invasive, diagnostic workup and delay both the correct diagnosis and the most appropriate therapy.

Although information of the reading habits of radiologists is scant, Andreisek et al[3] previously suggested that physicians focus on literature published in journals targeted to their respective specialties.[3] These authors compared 2 nearly identical, independently derived grading systems for spinal canal stenosis, one in the radiology literature and one in the orthopedic literature, as an example of duplicative and fragmented knowledge between the clinical and radiology literature. The findings of the present literature analysis indicate that it could be beneficial for radiologists to read, critique, and report imaging findings embedded in the nonradiology literature and to increase publication on these and similar CNS diseases within the radiology literature.

There are multiple potential explanations for the findings of this study. For example, clinical journals outnumber radiology journals. De-emphasis of case reports by radiology journals could also shift the balance of publication to clinical journals. Interestingly, most of included publications in the most common radiology journal in this study would not have been considered for publication in this journal after July 1, 2011, indicating some submissions since this date would presumably have been directed to other journals. Possibly in response to unmet demand for case report publication, several case report radiology journals have recently been introduced, but it is unclear if this will ultimately result in an increased number of radiology publications of uncommon CNS conditions. In addition, some publications containing imaging information potentially describe a new predisposing clinical condition, associated clinical symptoms or syndrome, treatment, affected demographic, or observed prognosis that might be most useful for a clinical audience. It is also possible that a single interested clinical specialist might see several patients with the same rare condition and publish his or her experience while the images of the individual patients are interpreted by several different radiologists. Additionally, lack of familiarity with conditions not reported in the radiologic literature may perpetuate a cycle wherein radiologists do not develop an interest in publishing on these uncommon conditions and remain unaware of new diagnostic issues. Other explanations include possible differences between neuroradiologists and neurologists in the degree of academic involvement, incentive to publish, and degree of subspecialization.

Although the Impact Factor ratings are not perfect measures of the value of an individual article or even a journal, the results of the current study indicate that the preponderance of articles on the selected topics in the clinical versus radiology literature is not explained by a lower clinical journal Impact Factor.[13] Radiologists were also authors in nearly half of articles within the clinical literature, indicating that lack of radiologist authorship should not account entirely for the proportional number of articles published within the clinical literature.

This literature analysis has several limitations. Only a limited number of conditions in a specified time period were evaluated, which could not capture all results from all potential search terms, search engines, neuroradiology diagnoses, or dates of publication. This study did not assess the actual rate of radiologist awareness of the imaging findings of the included conditions, references to the nonradiologic literature within radiology review articles, radiology textbooks, and/or radiology conferences. Rates of publication of imaging findings in nonradiology journals could differ with category or prevalence of a specific neuropathology and could differ across various subspecialty topics within radiology. A small number of articles were not included because of the lack of availability of all necessary information for data analysis. Finally, the Impact Factor analysis includes an assumption that articles in journals without listed Impact Factors can be represented.

CONCLUSIONS

This literature analysis demonstrates that uncommon conditions with potentially important CNS imaging findings are more often published in clinical rather than radiology journals, and radiologists are under-represented as authors on such publications. Radiologists should consider the use of broad literature searches and baseline journal reading to include clinical journals, as well as increasing the frequency of publication of imaging findings of uncommon conditions in the primary radiology literature.

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