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Short Communication

A scientometric and comparative study of rhabdomyosarcoma research by pediatricians and stomatologists

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Abstract *Background/purpose:* Rhabdomyosarcoma (RMS) frequently affects pediatric patients and common occurs in the head and neck region. The purpose of this study was to compare the scientometric characteristics of RMS publications by pediatricians and stomatologists.
Materials and methods: All the papers on RMS were comprehensively retrieved from the Scopus database, and divided into pediatricians and stomatologists groups.
Results: There were 2211 and 234 papers on RMS were published by rheumatologists and stomatologists, respectively. The total citation count was 63,868 and the *h* index was 111 for RMS publications by pediatricians, while the respective values for stomatologists were 2952 and 29. The two major subtypes of RMS are embryonal and alveolar types, and genetic alterations further categorize RMS into *PAX3/7–FOXO1* fusion-positive and fusion-negative types. Cancer chemotherapy mainly vincristine, dactinomycin, and cyclophosphamide, surgery, radiotherapy, and brachytherapy were the common keywords of treatment. The more common keywords such as age, facial neoplasms, mouth neoplasms, orbital neoplasms, gingiva, mandible, DNA-binding proteins, gene silencing, and Rh30 cell line were reported by stomatologists.
Conclusion: This study is the first comprehensive report of the scientometric characteristics of

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RMS publications by pediatricians and stomatologists, highlighting the need for increased awareness among clinicians to avoid diagnostic delays and ensure timely treatment.

Introduction

Rhabdomyosarcoma (RMS), as the most common soft-tissue sarcoma in children and adolescents, represents ~50 % of pediatric soft-tissue sarcoma.[1] Its most common site of occurrence are the head and neck regions (~40 %), followed by the genitourinary tract and the extremities.[2] Histologically, most of the pediatric cases involve embryonal (~70 %) and alveolar (~20 %) types, and a tiny fraction of them are spindle cell/sclerosing and pleomorphic variants.[1,2] Clinically, they are distinguished by differences in histopathology, genetics, and clinical presentation and outcome. Current treatment for RMS relies on chemotherapy, with surgery and radiation as adjunct therapies.[3,4] The treatment and prognosis for this sarcoma depends strongly on tumor size, location, tumor staging, histopathology, and child's age.[5,6] Owing to the use of these multimodal therapies, the cure rate in children can be improved to over 70 %; however, there is a poor prognosis for advanced cases with a 5-year survival rate of ~30 %.[7]

Generally, the diagnosis and treatment of RMS in children are conducted by pediatricians. Early-stage RMS signs, such as facial pain, swelling, loose teeth, or numbness or a feeling of heaviness in oral and maxillofacial regions, can be detected by stomatologists.[8] Hence, it is important to highlight the need for increased awareness among stomatologists to avoid diagnostic delays and ensure timely treatment. Decades of basic research and clinical studies developed by international collaborative groups have contributed to a better understanding of RMS pathophysiology and helped optimize clinical care. However, challenges remain despite improvements in RMS treatment and prognosis.[5—7] There were bibliometric/scientometric studies on some aspects of sarcomas,[9—11] with no relevant analysis of RMS. Hence, the purpose of the current study was to investigate and compare the scientometric characteristics and trends of RMS publications by pediatricians and stomatologists, in order to improve the early detection and management of this sarcoma and refer patients to adequate treatment.

Materials and methods

A literature search utilized the Scopus database, as done in previous bibliometric analyses.[12—14] Medical subject term "rhabdomyosarcoma" in the Title was used in the literature search to retrieve all the papers on RMS, without restriction to paper type and year of publication. Only English language literature was included because it is an international language of knowledge exchange. In clinical practice, pediatricians and stomatologists generally belong to the dermatology and stomatology affiliation, respectively. Hence, the papers with the word ("paediatri* OR pediatri*") and (dent* OR oral OR stomatolog*) in the affiliation generally represent scientific output of pediatricians and stomatologists, respectively. Accordingly, respective RMS-related publications by pediatricians and stomatologists were retrieved. The scientometric characteristics of all the eligible articles were reviewed. The following information was recorded as follows: publication year, title, abstract, keywords, citation count, paper type, journal, authorship, affiliation, and country/region of origin. Data search and extraction were performed independently by two investigators, and discrepancy of results was resolved in a consensus symposium. Microsoft Office Excel 365 was used for index model building, and the Bibliometrix Biblioshiny R-package software was used for bibliometric statistics. In this descriptive study, variables were presented as numbers and percentages. No comparisons were made, and thus no *P*-values were set.

Results

Citation characteristics

With the search strategy algorithm, 7152 papers on RMS were published until the time of the search. A total of 2211 (30.9 %) and 234 (3.3 %) papers were published by pediatricians and stomatologists, respectively (Fig. 1A). The total citation count was 63,868 and the *h* index was 111 for RMS publications by pediatricians, while the respective values for stomatologists were 2952 and 29. The detailed information on publication year, title, journal, citation count, authors, affiliation, abstract, keywords, and document types of the top-200 most-cited papers on RMS by pediatricians and stomatologists are presented in supplementary Table S1. To further clarify the treads of scientific output concerning RMS, we assessed the annual number and accumulated citation count of the papers during 2005—2024 (Fig. 1B). The annual number of the publications by pediatricians spirally changed from 31 to 127 during 2005—2024. The accumulated citations of the papers stably increased from 987 to 4413 during this period. The annual number of the publications by stomatologists slightly increased from 1 to 20 during 2005—2024, and the accumulated citations of the papers increased from 26 to 302 during this period.

Bibliometric characteristics

Fig. 1C displays cloud graphs of journal of publications, contributing authors, institutions, and countries/regions of origin. As for RMS publications by pediatricians, the journal of publication, contributing author, institution and country of origin with largest number of papers was *Pediatric Blood*

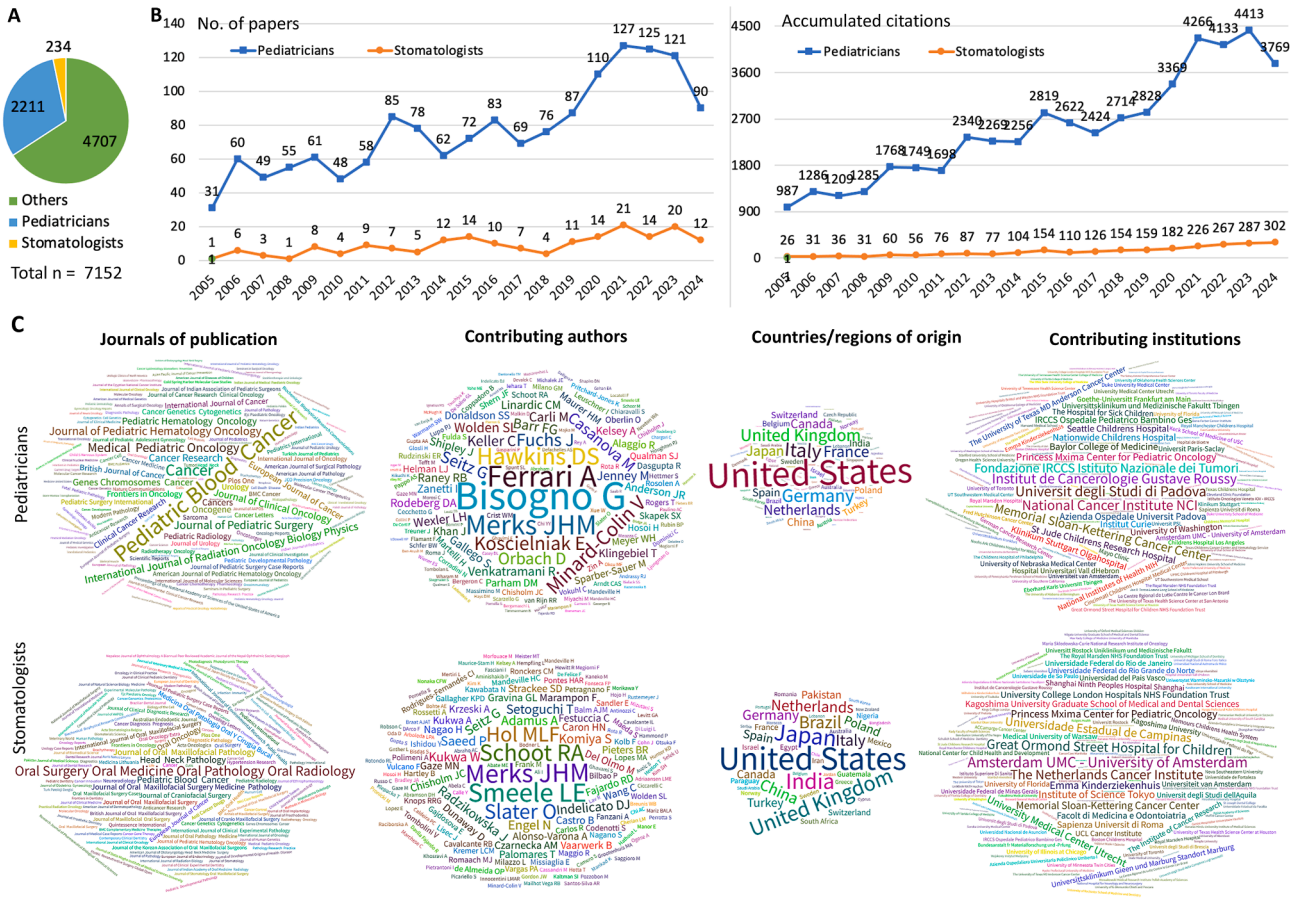

Figure 1 Bibliometric characteristics of RMS publications by pediatricians and stomatologists. (**A**) The numbers of the papers on RMS. (**B**) The annual number and accumulated citations of the papers during 2005—2024. (**C**) Cloud graphs of journal of publication, contributing authors, countries and institutions of origin regarding furcation involvement publications. The font size indicates the number of papers; a larger size means more papers in the cloud graphs.

& Cancer (n = 123), Bisogno, G. (n = 5), Università degli Studi di Padova (n = 121) and United States (n = 942), respectively. As for RMS publications by stomatologists, the journal of publication, contributing author, institution and country of origin with maximum number was *Oral Surgery Oral Medicine Oral Pathology Oral Radiology* (n = 16), Merks JHM and Smeele LE (both n = 10), Amsterdam UMC - University of Amsterdam (n = 10) and United States (n = 62), respectively. Supplementary Table S2 presents the journal of publication, contributing authors, institutions, and countries/regions with largest number of articles (rank, 1—10).

Research characteristics

Based on the frequency of keywords in all included papers, we highlighted the analysis of research characteristics of the papers on RMS by pediatricians and stomatologists. The study design, treatment keywords, common keywords used by pediatricians and stomatologists were identified. The most study design by pediatricians and stomatologists was controlled study and case report, respectively (Fig. 2A). Cancer chemotherapy mainly vincristine, dactinomycin, and cyclophosphamide, surgery, radiotherapy, and brachytherapy were the common keywords of treatment by pediatricians and stomatologists (Fig. 2B). Common keywords, such as child, preschool child, adolescent, pathology, genetics, immunohistochemistry, real time polymerase chain reaction (RT-PCR), embryonal rhabdomyosarcoma, alveolar rhabdomyosarcoma, prognosis, overall survival, transcription factor PAX3, cancer recurrence, metastasis, cell proliferation, metabolism, head and neck neoplasms, diagnostic imaging, gene mutation, Myod1 protein, and tumor marker were similar in the publications by pediatricians and stomatologists (Fig. 2C).

To elucidate the respective research directions and concerned topics, we further analyzed the distinctive keywords of the papers on RMS by pediatricians and stomatologists (Fig. 2D). Concerning clinical aspect, clinical trial, cohort analysis, in vivo study, cancer localization, cancer regression, newborn infant, echography, clinical outcome, disease-free survival, event-free survival, risk assessment, risk factor, and lymph node metastasis were distinctive keywords for pediatricians. Drug efficacy, carboplatin, epirubicin, topotecan, irinotecan, and treatment response were mainly mentioned by pediatricians. Concerning laboratory aspect, gene fusion, fluorescence in situ hybridization (FISH), western blotting, protein p53, transcription factor FKHR (FOXO1A) and PAX7 were distinctive

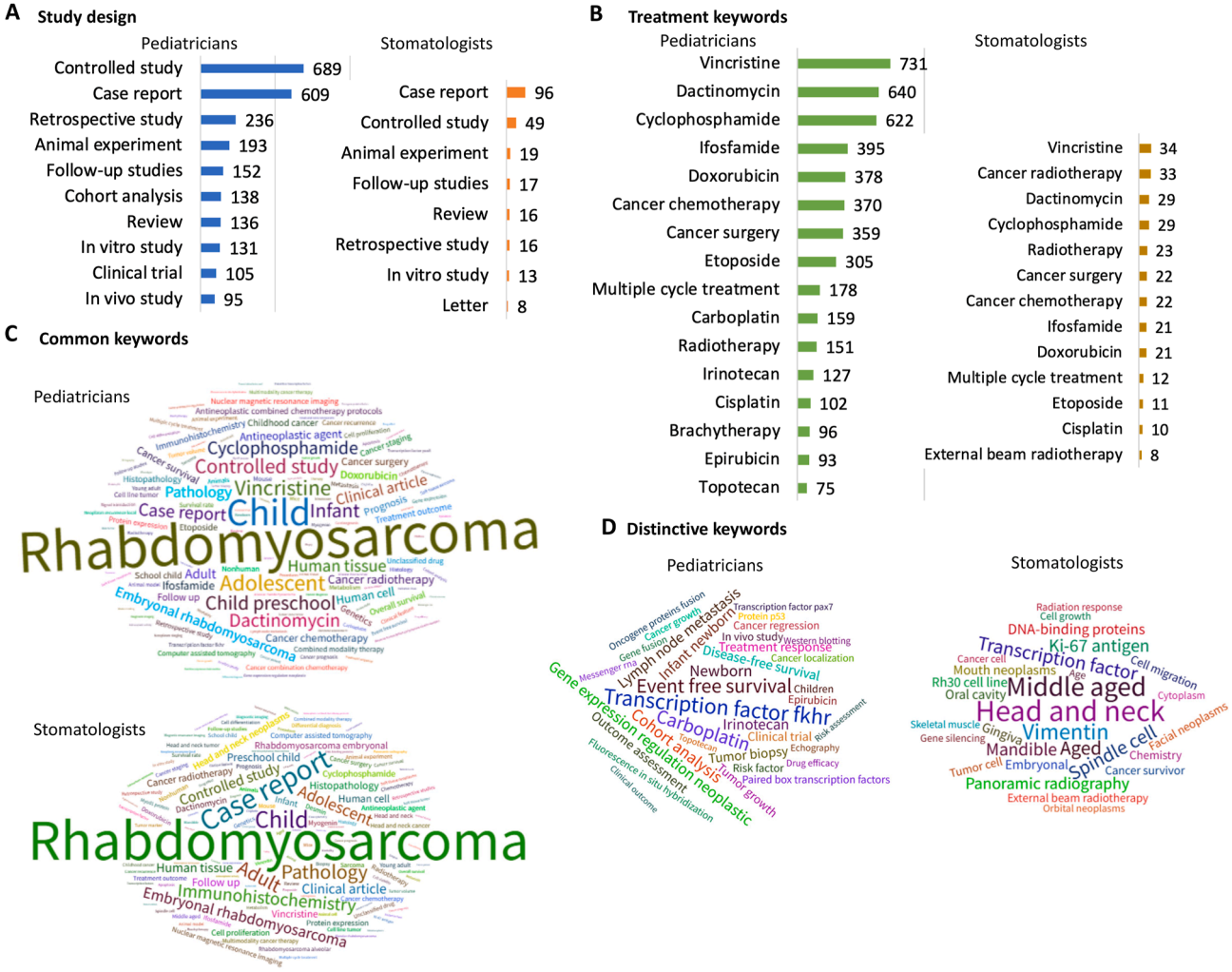

Figure 2 Research characteristics of RMS publications by pediatricians and stomatologists. (**A**) Study design. (**B**) Treatment keywords. Cloud graphs of (**C**) common keywords and (**D**) distinctive keywords. The font size indicates the number of papers; a larger size means more papers in the cloud graphs.

keywords for pediatricians. For the papers on RMS by stomatologists, age, facial neoplasms, mouth neoplasms, orbital neoplasms, gingiva, mandible, oral cavity, panoramic radiography, skeletal muscle, external beam radiotherapy, and radiation response were distinctive keywords of clinical aspect. Tumor cell, Rh30 cell line, spindle cell, cell growth, cell migration, DNA-binding proteins, gene silencing, Ki-67 antigen, and vimentin were distinctive keywords of laboratory aspect.

Discussion

RMS in children occurring in oral and maxillofacial region is commonly misdiagnosed, partly due to it being a rare malignant tumor. An earlier review article published in *Journal of Dental Research* highlighted the advances and prospective views in understanding the heterogeneity in childhood RMS.[15] This scientometric study attempted to analyze the bibliometric and research characteristics of RMS publications by pediatricians and stomatologists. Bibliometric characteristics identified in sequence would aid clinicians and researchers in choosing target journals, finding potential collaborators or partner institutions, as well as promoting mutual understanding and more reciprocal cooperation regarding RMS research.

The increasing numbers and citations of these papers each year suggest that childhood RMS has governed increasing attention and investigation. Although RMS is well-known to pediatricians, familiarity of its manifestations is essential among primary care practitioners and other concerned specialists including stomatologists. It is a remarkable fact that the RMS publications by the pediatricians were from the various regions (not only from head neck or maxillofacial region) of the whole body, whilst those RMS publications by stomatologists were from head and neck (maxillofacial) region only under normal conditions. The scale and citations of pediatricians' publications greatly outweigh stomatologists' ones, suggesting stomatologists can learn from and more cooperate with pediatricians regarding RMS research.

To comprehend the research directions and concerned topics of RMS, the research keywords of the publications by pediatricians and stomatologists were analyzed. The two major subtypes of RMS are embryonal (ERMS) and alveolar (ARMS) types that are distinct in their morphology and genetic alterations. In general, ERMS has a more favorable outcome, whereas ARMS has a worse outcome because of its aggressive and metastatic nature.[3–5] Oral and maxillofacial region is a challenging location for surgical resection given the potential for disfigurement and functional impairment. Primary surgery is possible for small tumors. Contrariwise, the preferred treatment is typically biopsy followed by chemotherapy and radiotherapy.[3,4] Vincristine, dactinomycin, and cyclophosphamide are the common drugs of RMS chemotherapy. Besides, brachytherapy offers conformal treatment to the target tissue while sparing healthy structures, and is therefore associated with less morbidity.

The transcription factors FKHR (*FOXO1A*, chr.13) and *PAX3* (chr.2)/*PAX7* (chr.1) and Myod1 and p53 proteins are the keywords regarding genetic and molecular alterations of RMS. The genetic alterations further categorize RMS into *PAX3/7—FOXO1* fusion-positive and fusion-negative types, as well as spindle cell/sclerosing RMS with *MYOD1* and *TP53* mutations. Approximately 80 % of tumors that are morphologically ARMS carry a *PAX3/7—FOXO1* fusion associated with poor outcome, whereas over 95 % of tumors that are morphologically ERMS have no *PAX3/7—FOXO1* fusion.[5–7] Accordingly, detection of a FOXO1 gene fusion has become a commonplace diagnostic and prognostic tool allowing tumors to be treated. Identifying FOXO1 gene fusion in RMS typically involves RT-PCR and FISH, and immunohistochemical assays using surrogate markers also offer a feasible and cost-effective option.

In summary, this study is the first comprehensive report of the scientometric characteristics of RMS publications by pediatricians and stomatologists, highlighting the need for increased awareness among clinicians to avoid diagnostic delays and ensure timely treatment.[16–18] Finding the scientometrics would elucidate the comprehensive identification and recognition of the important and relevant research topics concerned, but also help in improving in reciprocal collaboration and communication for investigations on this sarcoma.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jds.2025.09.019.

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