**Background:** With the global assimilation of “publish and perish” culture into institutional academics, there has been an exponential rise in the publication numbers. There are ~2500 PUBMED entries related to “anorectal malformation (ARM).” The young clinician in his pursuit to translate experimental research to bedside often finds himself lost “in the midst of plenty.” This bibliometric analysis has been conducted to codify the seminal work on ARM for future reference and pay tribute to the most impactful articles.

**Materials and Methods:** Thomson Reuters Web of Science citation indexing database and research platform was used to retrieve the most cited articles in ARM using appropriate search strings. The characteristics (name of authors, the total number of authors, the title of publication, journal of publication, year of publication, etc.,) of the 50 top-cited articles were analyzed.

**Results:** The analysis revealed that the Journal of Paediatric Surgery was leading the choice of journal for publication. While most of the publications originated from the United States of America, Alberto Pena was the most influential author. The most studied topic was on associated malformations, and the most common study design was cohort studies.

**Conclusion:** The approach of citation analysis provided us an opportunity to retrieve the most influential articles on ARM. The trends in research in ARM have also been analyzed, spreading over five decades.

**Keywords:** Anorectal malformation, bibliometric analysis, citations, Web of Science

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**INTRODUCTION**

Anorectal malformations (ARMs) encompass a variety of congenital malformations affecting the distal anus and rectum of both boys and girls. Other associated anomalies can be seen in the urinary and genitourinary tracts as well as several other organ systems. The incidence is approximately 1 in 5000 live births. Several approaches and surgical techniques were propounded that continued to get refined over the subsequent centuries till the 1980s when the posterior sagittal approach was put forward by Peña and DeVries.[1] Over the years, better anatomical delineation, a better understanding of the anatomy and physiology of the anorectal region at birth have standardized the diagnosis and initial management of neonates with ARM.

The concept of bibliographical analysis helps us understand the influence of these papers on the research landscape and provides us with an insight into the most influential papers published. The process involves examining the citation history of an individual publication and obtaining additional quantitative data.

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on different aspects of the paper such as the authors, institute, and country of origin of the research. Many surgical specialties such as general surgery, plastic surgery, trauma, orthopedic surgery, cardiothoracic surgery, and even pediatric surgery have deployed bibliometric analysis in different dimensions. However, the territory of ARM remains unexplored on this front.

To the best of the authors’ knowledge, this is the first study trying to explore the characteristics of the top 50 cited articles published on the subject over the past 50 years (1969–2019). The current analysis has been conducted to codify the seminal work on ARM for future reference and pay tribute to the most impactful articles. The analysis is also expected to be considered as a reference for the most influential papers on ARM during the study period in years to come.

**Materials and Methods**

Thomson Reuters Web of Science citation indexing database and research platform (Web of Science core collection database maintained by Clarivate Analytics) was used to conduct the search. The “Topic” field and the “Title” field were searched using the search string “ARM OR anorectal atresia OR anal atresia OR imperforate anus” with a filter applied to include publications from January 01, 1970, to December 31, 2019. The dataset was further filtered to include only English language manuscripts and those indexed under the Science Citation Index Expanded, Conference Proceedings Citation Index, or Emerging Sources Citation Index. Articles thus obtained were reviewed, and the data entered into an Excel spreadsheet. The process of data retrieval and data entry was completed in 24 hours, and the master chart was frozen on May 10, 2021. The various parameters recorded were the name of authors, the total number of authors, the title of publication, journal of publication, year of publication, country and the institute of the first author, and the type of paper. The quality of the articles was gauged by the total number of citations and the average number of citations received per year (calculated by the number of total citations by the number of years since publication). Among multi-institutional studies and studies with more than six authors, the first six authors only were considered for analysis. For this study, we categorized the papers as original papers (basic science/animal study/clinical study/cohort studies/retrospective comparative study), review article, and case report or small case series. Subgroup analysis was done between collaborative articles and single-institution articles and articles published in pediatric surgery-specific journals and those published in nonpediatric surgery-specific journals.

**Statistical analysis**

The summary information (journal, year of publication, number of citations, and the average citations received per year) of the top 50 most cited articles in ARM is depicted in Table 1. The detailed analysis of the top 50 most cited articles in ARM with regard to the journal of publication and the citations received is provided in Table 2. The bivariate distribution of the 50 most cited article was assessed in terms of three parameters across the pediatric-specific journal and nonpediatric-specific journals [Table 3]. These three parameters were number of articles, total citations, and the citations per article. The test of significant difference in the proportion of articles and citations was assessed by the Z-test for two proportions. The last parameter (citation per article) was a ratio statistic. The null hypothesis under this scenario was that the rate ratio of citation across two specialties was equal to one against the alternative hypothesis that it is different from one. This was assessed by the help of a suitable R command by declaring the total variable citations and the total cited article as the vector and applying the exact rate ratio test, assuming Poisson count. A $P < 0.05$ was considered to be statistically significant. Data entry was done using Microsoft Excel®, and statistical analysis was performed using licensed Stata/SE version 12.0. Institutional review board approval was not sought, as the database that was being analyzed was already available in the electronic form in the public domain.

**Results**

A total of 1510 articles were retrieved in the Web of Science search with a total no of 18,320 citations (average 11.42 citation/article) on these articles. After arranging these 1510 articles in descending order of citations received, the top 50 articles were considered for further analysis. These top 50 articles are described in with their corresponding citation counts [Table 1].

These 50 articles received a total of 4796 citations (average 47.96 citations/article). The maximum citation count registered was 470, and the minimum noted was 54. When the average number of citations received per year was considered, the top 3 rankings were secured by the article numbers 2, 4, and 1, respectively [Table 1].

These top 50 articles were published between the year 1969 and 2011. These 50 articles were published in 21 journals and are illustrated in Table 2. Out of these 21 journals, the journal with the highest number of articles in the ranking of the 50 top-cited articles was the “Journal of Pediatric surgery” (JPS) ($n = 26, 2237$ citations). The second rank was secured by the “American Journal of
| Title                                                                 | Journal                        | Year of publication | Number of citations | Average citations received/year |
|----------------------------------------------------------------------|--------------------------------|---------------------|---------------------|----------------------------------|
| VATER association – vertebral defects, anal atresia, T-E fistula with EA, radial and renal dysplasia – spectrum of associated defects | Journal of Pediatrics           | 1973                | 470                 | 10                               |
| Preliminary report on the international conference for the development of standards for the treatment of ARMs | JPS                            | 2005                | 309                 | 20.6                             |
| Congenital hypothalamic hamartoblastoma, hypopituitarism, imperforate anus, and postaxial polydactyly – a new syndrome 1. Clinical, causal, and pathogenetic considerations | American Journal of Medical Genetics | 1980                | 236                 | 5.9                              |
| ARMss                                                                 | Orphanet Journal of Rare Diseases | 2007                | 213                 | 16.3                             |
| Laparoscopically assisted anorectal pull through for high imperforate anus – A new technique | JPS                            | 2000                | 184                 | 9.2                              |
| ARMs caused by defects in sonic hedgehog signaling                     | American Journal of Pathology   | 2001                | 164                 | 8.6                              |
| Advances in the management of ARMs                                     | American Journal of Surgery     | 2000                | 134                 | 6.7                              |
| Is normal bowel function possible after repair of intermediate and high anorectal malformations | JPS                            | 1995                | 133                 | 5.3                              |
| Outcomes from the correction of ARMs                                  | Current Opinion in Pediatrics    | 2005                | 118                 | 7.8                              |
| Hereditary syndrome of imperforate anus with hand, foot, and ear anomalies | Journal of Pediatrics            | 1972                | 110                 | 2.2                              |
| Quality of life for children with fecal incontinence after surgically corrected ARM | JPS                            | 2000                | 103                 | 5.1                              |
| Bowel management for fecal incontinence in patients with ARMs         | JPS                            | 1998                | 95                  | 4.3                              |
| Anorectal malformations – evaluation of associated spinal dysraphic syndromes | JPS                            | 1988                | 93                  | 2.9                              |
| Fecal continence and quality of life for adult patients with an operated high or intermediate ARM | JPS                            | 1994                | 89                  | 3.4                              |
| Associated malformations in patients with anorectal anomalies         | European Journal of Medical Genetics | 2007                | 85                  | 6.5                              |
| PSARP – results in the management of 332 cases of ARMs                | Pediatric Surgery International | 1988                | 85                  | 2.6                              |
| The tethered spinal cord in patients with ARMs                        | JPS                            | 1997                | 81                  | 3.5                              |
| Colostomy in ARMs: A procedure with serious but preventable complications | JPS                            | 2006                | 77                  | 5.5                              |
| Currarino triad: ARM, sacral bony abnormality, and presacral mass – A review of 11 cases | JPS                            | 1997                | 77                  | 3.3                              |
| Analysis of 1992 patients with ARMs over the past two decades in Japan | JPS                            | 1999                | 76                  | 3.6                              |
| Cloaca, the most severe degree of imperforate anus – Experience with 195 cases | Annals of Surgery               | 1998                | 76                  | 3.4                              |
| OEIS complex: A review of 14 cases                                   | American Journal of Medical Genetics | 2001                | 73                  | 3.8                              |
| Congenital hypothalamic hamartoblastoma, hypopituitarism, imperforate anus, and postaxial polydactyly – a new syndrome 2. Neuropathological considerations | American Journal of Medical Genetics | 1980                | 71                  | 1.7                              |
| One hundred three consecutive patients with ARMs and their associated anomalies | Archives of Pediatrics and Adolescent Medicine | 2001                | 70                  | 3.6                              |
| Urologic injuries associated with repair of ARMs in male patients     | JPS                            | 2002                | 68                  | 3.7                              |
| New mouse models of congenital ARMs                                  | JPS                            | 2000                | 68                  | 3.4                              |
| The OEIS complex (OEIS) – recurrence in sibs                         | Journal of Medical Genetics      | 1992                | 67                  | 2.3                              |
| Fecal continence and quality of life in adult patients with an operated low ARM | JPS                            | 1992                | 66                  | 2.3                              |

Contd...
Medical Genetics” (n = 3, 380 citations). There were two journals in the third place (archives of diseases in childhood and journal of pediatrics) with two papers each and 121 and 580 citations, respectively. In addition, there were 19 other journals that had at least one article in the list of the 50 top-cited articles [Table 2].

There were three journals that were pediatric surgery specific (JPS, Pediatric surgery international and Seminars in pediatric surgery), accounting for 28 of the 50 articles (2380 citations, 49.6% of total citations received by the 50 articles); the rest 22 articles were published in nonpediatric surgery-specific journals (2416 citations, 50.4% of total citations received by the 50 articles). The comparative analysis between the pediatric surgery-specific journals and the nonpediatric surgery-specific journals is tabulated in Table 3.

The distribution of these fifty articles over the five decades of the study period is illustrated in Figure 1.
The most productive in terms of the most influential articles were published between 2000 and 2009 with 19/50 (38%); in contrast, only one article (2%) of the top 50 cited articles was published between 2010 and 2019, receiving 58 citations. Before the year 2000, a trend of gradual increase in the number of articles which found a place in the top 50 cited articles as well as their corresponding citations was noted from the year 1969 onward [Figure 1].

Maximum number of studies (n = 25, 2740 citations) were from the United States of America (USA), followed by Finland (n = 6, 461 citations), Netherlands (n = 5, 292 citations), and Canada (n = 3, 286 citations). The United Kingdom and Germany had two articles, each with 122 and 369 citations, respectively. Australia, Japan, Peoples Republic of China, South Korea, Saudi Arabia, and Switzerland were the country of origin of one article, each in the top 50 rankings with total citations of 526 citations. The number of articles having a contribution from more than one institute was 12. The number of institutes contributing to these 12 multi-institutional (collaborative) articles ranged from 2 to 21 institutes with a median of 3. Further analysis of these two categories of articles (single institution vs. collaborative) is projected in Table 4. It was observed that the total citations received were significantly more in the collaborative articles (P < 0.0001). Furthermore, a significant difference was observed in comparing the average citations per article between these subcategories of articles (P ≤ 0.0001).

The top three institutions according to the number of articles contributed to the top 50 articles were the University of Helsinki (n = 6, 461 citations), the University of Washington (n = 4, 688 citations), and the Hospital for sick children, Toronto (n = 3, 286 citations).

One hundred and ninety-three authors contributed to the top 50-cited articles. The number of authors in each article ranged from 1 to 26, with a median of 3. The top 3 contributors are Alberto Peña (n = 10, 1245 citations), Risto J Rintala (n = 8, 583 citations), and Harry Gustav Lindahl (n = 6, 468 citations).

The most cited topic was related to the “associated condition” in ARM. The number of papers on various topics on ARM and their citations is depicted in Table 5.

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**Table 2: Detailed analysis of the top 50 most cited articles in anorectal malformation with regards to the journal of publication and the citations received**

| Journal                                      | Number of articles | Citations received |
|----------------------------------------------|--------------------|--------------------|
| JPS                                          | 26                 | 2237               |
| American Journal of Medical Genetics         | 3                  | 380                |
| Archives of Disease in Childhood             | 2                  | 121                |
| Journal of Pediatrics                        | 2                  | 580                |
| American Journal of Pathology                | 1                  | 164                |
| American Journal of Human Genetics           | 1                  | 61                 |
| American Journal of Roentgenology            | 1                  | 66                 |
| American Journal of Surgery                  | 1                  | 134                |
| Annals of Surgery                            | 1                  | 76                 |
| Archives of Pediatrics and Adolescent Medicine | 1                | 70                 |
| Current Opinion in Pediatrics                | 1                  | 118                |
| Diseases of the Colon and Rectum             | 1                  | 60                 |
| European Journal of Medical genetics         | 1                  | 85                 |
| European Journal of Pediatrics               | 1                  | 54                 |
| Journal of Medical Genetics                  | 1                  | 67                 |
| Orphanet Journal of Rare Diseases            | 1                  | 213                |
| Pediatric Neurosurgery                       | 1                  | 56                 |
| Pediatric Radiology                          | 1                  | 54                 |
| Pediatric Surgery International              | 1                  | 85                 |
| Quality of Life Research                     | 1                  | 57                 |
| Seminars in Pediatric Surgery                | 1                  | 58                 |
| JPS: Journal of Pediatric Surgery            |                    |                    |

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**Table 3: The differences observed among the articles published in pediatric surgery specific journal or otherwise**

|                         | Pediatric surgery specific journal (3/21) | Nonpediatric surgery specific journals (18/21) | P       |
|-------------------------|------------------------------------------|----------------------------------------------|---------|
| Number of articles      | 28/50                                    | 22/50                                       | 0.23 (Z-test) |
| Total citations         | 2380/4796                                 | 2416/4796                                   | 0.47 (Fischer’s exact test) |
| Citations per article   | 85                                       | 109.8                                       | 0.02 (Fischer’s exact test) |

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**Figure 1:** A graphical description of the number of top 50 most cited articles in anorectal malformation articles and the citations received over various time frames of the study period
A note must be made of the total number of papers add up to more than 50 as the asterisked marked topics have been combinedly dealt with in some articles, which therefore have been accounted separately under each topic.

The distribution of the article based on the type is illustrated in Figure 2. The original experiments were further subcategorized into six types based on the research methodology used.

DISCUSSION

The number of citations an article receives showcases the interests of the researchers in that particular subject. Studying the frequently cited articles provides information on the important topics and the attention and growth in research a particular area has received. When the bibliometric analysis of the research pertaining to ARM was performed, it was noticed that the most frequently cited topic was on associated congenital anomalies. This pattern is expected since ARM can have a myriad of associated conditions. Any publication dealing with an ARM is bound to discuss this aspect of ARM in the manuscript and quote the findings of previous papers on this topic. The same argument holds true to explain the fact that the highest citation was received by a manuscript by Quan et al. titled “VATER association-vertebral defects, anal atresia, T-E fistula with esophageal atresia (EA), radial and renal dysplasia– spectrum of associated defects.”[7] This study is the original description of VATER association and since then has not only gathered the highest citation in ARM (n = 470) but also topped the rankings in a bibliometric analysis conducted on EA by Feng et al.[8]

The classification of ARM has also changed as the understanding of this complex malformation got refined. The basis of uniform documentation of any case of ARM is a proper categorization of the defect per se only when uniform documentation and categorization has achieved the results of a particular technique or a treating center can be compared, and inferences are drawn. This led to the development of the “Krickenbeck Classification” of ARM in 2005. The article contains the summary of 26 international experts in the ARM workshop, which has been the second-highest cited article in ARM to date.[9]

As far as the proportion of articles published in the pediatric surgery-specific journal or nonpediatric surgery-specific journal is concerned, the top 50 cited articles showed no difference. In contrast to the bibliometric study done by Feng et al. on EA, they observed that one-third of total articles on EA were in pediatric surgery-specific journals.[8] Furthermore, we did not observe any difference in the quality of articles between these two subcategories of articles as assessed by the citations received per article in the two categories. This is in sharp contrast to the findings of Feng et al., as they had noted that the highest citations received per article were from the articles that were published in high-impact nonpediatric surgery journals.[8] Their speculation was that the authors of original articles bearing significant research work and results on EA probably preferred to submit their results in high-impact journals with a wider audience and specialties rather than in journals directed to only the pediatric surgery community. Unlike their speculation, our findings highlight that ARM per se is a condition which is mainly managed by pediatric surgeons, in comparison to the multimodality approach in EA where the role of a pediatrician/neonatology is significant, and therefore, influential research work in EA could find a place in nonpediatric surgery-specific journals also. Furthermore, one can note that the citations and the citations per article received [Table 3] in the pediatric surgery-specific journals are marginally less than that of nonpediatric surgery journals. One must not, therefore, consider the papers in pediatric surgery journals being inferior to those of nonpediatric surgery journals; rather, it may be the phenomena of a larger audience and wider dispersion of the articles in nonpediatric surgery journal playing a role in accruing higher citations.

The time period between 2000 and 2009 saw the publication of the greatest number of influential articles in ARM. This finding is explainable since research in ARM and pediatric surgery has grown exponentially, and every new research work has a tendency to quote the recent most relevant article in the field. The older articles are generally not the first choice unless the author does not have an appropriate option to choose. The effect of duration of exposure of an article to the scientific community upon the citation count received

| Table 4: The differences observed among the articles published by a single institute as compared to collaborative articles |
|---------------------------------------------------------------|
| **Single institution articles** | **Collaborative articles** | **P**          |
| Number of articles | 38/50 | 12/50 | <0.0001 (Z test) |
| Total citations | 3367/4796 | 1429/4796 | <0.0001 (Fisher’s exact test) |
| Citations per article | 88.6 | 119 | <0.0001 (Fisher’s exact test) |
Dhua, et al.: Bibliometric study in anorectal malformation

is also exemplified by the figures obtained between the time period 2010 and 2019, which is the lowest.

From our findings, it would not be wrong to state that pioneering research work in ARM has originated from institutes in the USA, followed by centers in Europe (Finland and Netherlands) and Canada. The highest number of articles in the list of top 50 was published in the JPS. It is noteworthy to note that the JPS is the official journal of several professional societies (Section on Surgery of the American Academy of Pediatrics, the British Association of Paediatric Surgeons, the American Pediatric Surgical Association, the Canadian Association of Paediatric Surgeons, and the Pacific Association of Pediatric Surgeons). Therefore, it is easy to assimilate the conditions leading JPS to be receiving high-standard articles and subsequently publishing the highest number of influential papers in ARM. When we compared single-institution authored articles and those that had collaboration from multiple institutions, it was noted that the number of articles, total citations, and citations received per article were significantly more among the collaborative articles.

Feng et al. in their bibliometric study on EA also noted that half of the best-connected authors were responsible for the most productive and most cited authors in their study. This leads to prove that a collaborative research and publication have higher chances of adding value to the paper as far as receiving higher citations is concerned and can become relatively more influential compared to those articles which are authored by single institution only.

Among the authors, the top names are Alberto Pena, Risto J Rintala, and Harry Gustav Lindahl. While Pena A (USA) revolutionized the surgical management of ARM by introducing the concept of posterior sagittal anorectoplasty, Rintala RJ, and Lindahl GH’s articles predominantly emphasized the long-term outcomes of patients of ARM by longitudinal follow-up of a large cohort of ARM patients even up to their adulthood.

The most common topic among the most cited articles in ARM was “associated conditions” in ARM followed closely by “outcome.” The outcome of management is what we look forward to as surgeons. As mentioned before, the surgical reconstructive technique has evolved

**Table 5: Detailed analysis of the topic of research of the top 50 most cited articles in anorectal malformation**

| Topic                                | Number of articles | Citations received |
|--------------------------------------|--------------------|--------------------|
| Associated condition                 | 14                 | 1404               |
| Outcome*                             | 15                 | 1117               |
| Pathology and genetics*              | 5                  | 594                |
| Surgical management*                 | 5                  | 546                |
| QOL                                  | 6                  | 438                |
| Developing standards for an          | 1                  | 309                |
| International Classification of ARM  |                    |                    |
| Laparoscopy                          | 1                  | 184                |
| OEIS complex                         | 2                  | 140                |
| Cloaca*                              | 1                  | 76                 |
| Antenatal evaluation                 | 1                  | 66                 |
| Anatomy                              | 1                  | 60                 |
| Enteric nervous system               | 1                  | 57                 |
| Anorectal manometry                  | 1                  | 57                 |
| The practice of ARM Surgery - Survey| 1                  | 55                 |

*Indicates that the particular article/ articles under this head also dealt with other topics listed in the table and has been counted under those heads also, hence, the total number of articles add to more than 50, ARM: Anorectal malformation, QOL: Quality of Life, OEIS: Omphalocele-extrophy-imperforate anus-spinal defects
over decades, and the acceptance of any particular technique is bound to happen when the results of such a technique are showcased, discussed, endorsed by all, and modified as required. Long-term outcomes and quality of life-related topics are also important since the surgery is performed in a child who has an entire life span ahead of him. These longitudinal “outcome” results provide a very important facet of the surgical management of ARM as the child becomes an adult. Such “outcome-“based papers are therefore bound to get repeated citations in the newer publications dealing with ARM. The etiology and molecular mechanisms for aberrations leading to the occurrence of ARM in the fetus are also the focus of study, as evidenced by five articles among the rankings. As laparoscopy has also become an accepted modality of surgical treatment for select cases of ARM, more and more articles are being published with modification, early and long-term results of the minimally invasive surgery for the management of ARM. Such studies must have cited Georgeson et al. for quoting the first description of laparoscopy in ARM, and therefore, this article has found a place among the top 50 cited articles.[10] It was notable that an important area of research on ARM, i.e., animal models did not find a place in our list. A similar observation was also made by Feng et al. in their study of top 100 cited articles on EA where only two articles on animal models were present in their listing.[9]

A significant proportion of articles in this list of top-cited articles is retrospective studies on a cohort of patients. It was striking to note that meta-analysis and systematic reviews, which form the top rank in the pyramid of the level of evidence, are lacking in this list. This could be explained by the fact that it is technically difficult to design a prospective randomized controlled trial in a rare disease in which a lot of anatomical variation and heterogeneity exists among different patients. Hence, randomized studies in ARM are far and few, and therefore, articles based on meta-analysis and randomized control trials are underrepresented in our study.[11,12]

There are some limitations to this analysis that merits discussion. The citation count is a dynamic value, and hence, our analysis is akin to taking a snapshot which is bound to get modified over time as research continues to evolve. One may also note that a citation count does not reflect whether it is a citation made in a positive or a negative context in a particular article.[13] It is also known that the type of article also matters; for example, meta-analyses or systematic reviews tend to gather more citations than others.[14] Articles published earlier get more exposure to the scientific community and therefore have more probability of accumulating higher citations than those which are published recently. The “Web of Science” citation numbers might not be wholly representative of the true citation count as many available manuscripts may not have been included under its purview. The utilization of multiple databases can generate a higher citation number.[6] Several biases such as institutional bias, self-citation, powerful person, and language bias have not been factored in this study, and these could have affected the manuscript’s rankings. Finally, only the first six authors were considered for analysis, and publications that were coauthored by more than six authors may appear to be under-represented by our study design.

CONCLUSION
This bibliometric analysis is the first in the field of pediatric surgery to provide a list of the top 50 cited articles dedicated to ARM. Through this study, the research areas and major academic publications pertaining to ARM were identified. This analysis of the citation count would provide a reference for quoting the most influential papers in the field of ARM in future publications.

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Conflicts of interest
There are no conflicts of interest.

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