Singaporean Journal of Scientific Research(SJSR)
An International Journal (AIJ)
Vol.18.No.1 2026,Pp.1-7
ISSN: 1205-2421
available at :www.sjsronline.com
Paper Received : 04-01-2026 Paper Accepted: 01-03-2026
Paper Reviewed by: 1.Prof. S.Subramaniam Yu 2. Dr.K. Rajakumaran
Editor : Dr. Chen Du Fensidal

SCIENTOMETRICS ANALYSIS ON PUBLICATIONS TRENDS OF OLIGOSACCHARIDOSES

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Abstracts
Scientometrics or bibliometrics is a tool to analyze scientific research trends and impact. Oligosaccharidoses are rare lysosomal storage disorders caused by enzyme deficiencies, leading to the buildup and excretion of undegraded substrates. We diagnosed about 40 cases of oligosaccharidoses in Malaysia. Despite increasing research in this area, no bibliometric study has been conducted on oligosaccharidoses. This study aims to examine the distribution, trends, and impact of related scientific publications. The search term of keyword was "oligosaccharides". Data were retrieved from Google Scholar and Scopus, two widely used academic databases. A total of 739 articles were included in the final dataset. The cleaned data were then imported into VOSviewer. The most active authors were Sewell (12), Harvey David J (8), O' Brien John S (8), Lebrilla Carlito b (7), and Winchester Bryan (7). Single authorship was 68, two authors 106, three authors 97, four authors 93, five authors 75. Top publications of oligosaccharidoses in J. Inherit. Metab. Dis. (37), J. Biol. Chem. (31), Mol. Genet. Metab. (29), Clin. Chem (27), Glycobiology (26). Most frequent themes identified included inborn error (24), gangliosidosis (21), carbohydrate (17), milk oligosaccharides (13), Pompe disease (12). Highest publications were from United States (195), Japan (74), Germany (68), United Kingdom (50), The Netherlands (43), France (40) Italy (30), Canada (23), Australia (22), Brazil and Finland (18), Poland (15). This bibliometric analysis provides a comprehensive overview of the research on oligosaccharidoses and provide guide for future research.

Keywords: oligosaccharidoses, publications, authors, collaboration, themes.

1.Introduction

Scientists communicate through writing of scientific articles, books and other literature form to explain, argue, enthuse, and convince, at many different levels (Kelly, 2020). Scientometrics analysis have been studied for providing quantitative analysis of the published scientific

articles. Scientometrics was broadly known as "infometrics" (Egghe & Rousseau, 1990), identified as the quantitative methods in library, documentation and information science. Specifically, it was closely known as "webometrics" or "bibliometrics" (Ellegaard & Wallin, 2015). According to Pritchard (1969) who defined the term of bibliometrics as the application of mathematics and statistical methods to books and other communication media, Hulme (1923) who initiated the term "statistical bibliography". The term "Scientometrics", derived from the Russian 'naukometria', was the study of the measurement of scientific and technological progress (Egghe, 1988). The advancement of scientometrics progress in line with the technological improvements, whereas the recent new metrics included Download statistics, page ranks, bookmarking tools and sharing on social media (Ellegaard & Wallin, 2015). The data measured including authors, affiliations of authors, words/subjects frequencies, usage statistics such as citations (Wilson, 2012).

Oligosaccharidoses are group of lysosomal storage disorders due to specific enzymes deficiencies. There were about ten types of oligosaccharidoses such as fucosidosis, aspartylglucosaminuria, sialidosis, GM1-gangliosidosis, GM2-gangliosidosis, galactosialidosis, α-mannosidosis, β-mannosidosis, Pompe diseases, Schindler's diseases, and I-cell. The clinical spectrum of the diseases includes not only lysosomal storage disorders, but neurological symptoms, psychiatric symptoms in adults (Federico & Silvia, 2016). These disorders could be analysed by thin layer chromatography (TLC), fluorimetric (4-MUG) enzyme assay, radio-active enzyme assay for sialic acid disorder, HPLC, UPLC, tandem mass spectrometry (TMS), and molecular analyses.

There was no bibliometric analysis conducted on human oligosaccharidoses. Oligosaccharides are two to forty sugar units. Deficiencies of oligosaccharidoses enzymes will cause storage disorders and other pathological symptoms. Since there was no bibliometric analysis conducted on oligosaccharidoses, we proposed this analysis to be conducted in Malaysia.

2. Methodology

This retrospective study were conducted on all types of documents on oligosaccharidoses in the google scholar database until mid of this year. The publication data of title, author name, year, keyword, citations, issues, and page number were extracted from google scholar database. Random selection of 10% percent from the dataset were matched with the published articles in journals as a measure of "quality control". Data merging, pivoting and aggregation analysis were conducted by using Microsoft Excel and collaboration network graph of co-authorship and subject area/keywords clusters were prepared with the software VOSviewer and R-package. Collaboration coefficient (CC) was analysed. CC taking into account both the average number of authors per paper and the proportion of multi-authored papers. The CC typically falls between 0 and 1, with 0 representing purely single-authored papers and values closer to 1 indicating higher levels of collaboration.

3.Results

There were 737 publications searched and analysed. Types of publications were divided into mainly articles (277), and reviews (190), mini review (1), editorial (2), special article (1), original article (20), original paper (2), original research (1), original contribution (1), original investigation (3), regular article (2), regular papers (1), research report (1), report

(1), research article (9), research communication (2), research letter (1), case report (174), full length article (1), comparative study (22), ACMG technical standard (1), brief clinical report (1), brief communication (2), brief technical note (1), communication (1), short communication (5), clinical study (1), clinical trial (4), multicentre study (1), observational study (1), guide (1), letter (1), and supplement (1).

The longest number of articles pages was 223 pages reviews written by Harvey, David J on Analysis of carbohydrates and glycoconjugates by matrix-assisted laser desorption/ionization mass spectrometry: an update for 2019–2020 (2023). He also written same reviews with 139 pages: an update for 2013 – 2014 (2018), and 129 pages review: an update for 2007 – 2008 (2012) in Mass Spectrometry Reviews.

Most frequent themes: inborn error (24), gangliosidosis (21), carbohydrate (17), milk oligosaccharides (13), Pompe disease (12) in Fig. 1. Top publications of oligosaccharidoses

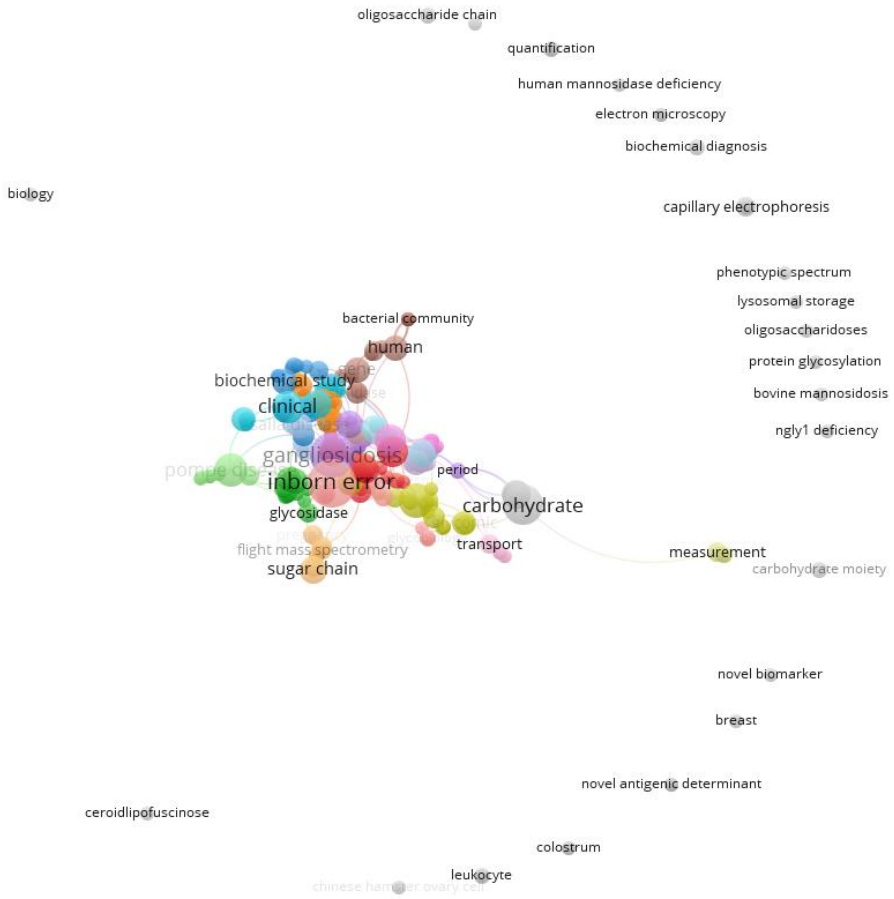

Fig 1: VosViewer on themes. High occurrence: inborn error (24), gangliosidosis (21), carbohydrate (17), milk oligosaccharides (13), Pompe disease (12).

were published in J. Inherit. Metab. Dis. (37), J. Biol. Chem. (31), Mol. Genet. Metab. (29), Clin. Chem (27), Glycobiology (26). Besides, most active authors were Sewell, Germany (12

occurences), Harvey David J, United Kingdom (8), O' Brien John S, United States (8), Lebrilla Carlito b, United States (7), and Winchester Bryan, United States (7) as shown in Fig. 2. Highest publications were from United States (195), Japan (74), Germany (68), United Kingdom (50), The Netherlands (43), France (40) Italy (30), Canada (23), Australia (22), Brazil, and Finland (18), Poland (15) as shown in Fig 3. These implicated that United States, Asian, European and Australian authors involved in oligosaccharidoses research and publications. Most papers were written in English. Two papers in Polish, one in Italy, one in Brazil, one in Germany and one in Spanish.

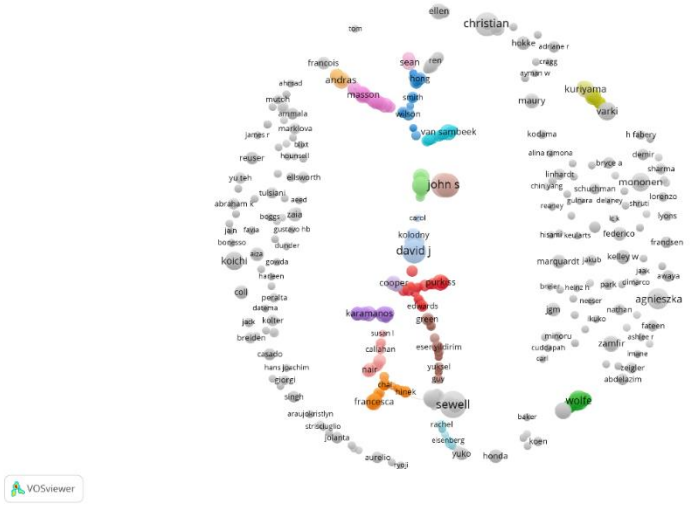

Fig 2: VosViewer on authors. Sewell (12 occurences), Harvey David J (8), O' Brien John S, (8), Lebrilla Carlito b, (7), and Winchester Bryan, (7).

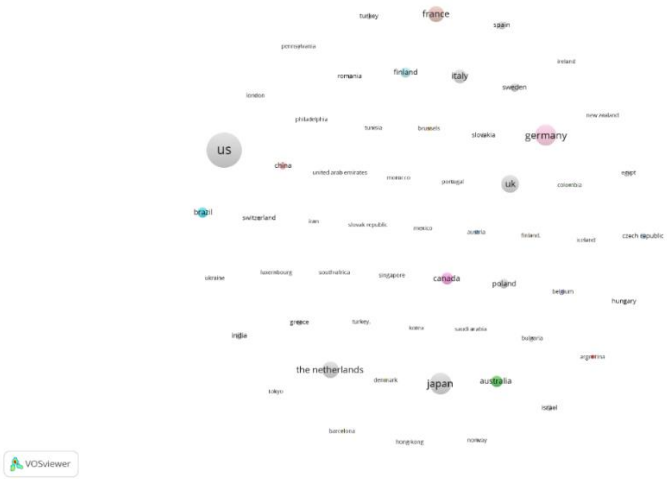

Fig 3: VosViewer on countries. United States (195), Japan (74), Germany (68), United Kingdom (50), The Netherlands (43), France (40) Italy (30), Canada (23), Australia (22), Brazil and Finland (18), Poland (15).

Number of articles with single authorship was 68, two authors 106, three authors 97, four authors 93, five authors 75. Degree of collaboration was 0.904 with average collaboration index of 4.86; and average collaboration coefficiency was 0.6795. Highest authorship was 17 authors with paper on N-acetylneuraminic storage with sialuria.

Most publications were found in the year of 1983 (29), followed by 2014 (26), both 2019 and 2020 (25); lowest publication was one paper annually in the year 1954, 1960, 1966 and 1969. First paper found was in the year of 1954 on "The raffinose family of oligosaccharides" in Advances in Carbohydrate Chemistry. Higher publications were found starting the year 1980 with publications > 10 papers with highest 1983 (29 papers). Afterwards, publications remained for > 10 papers yearly and rarely > 20 papers (in the year 1989, 2006, 2014, 2019).

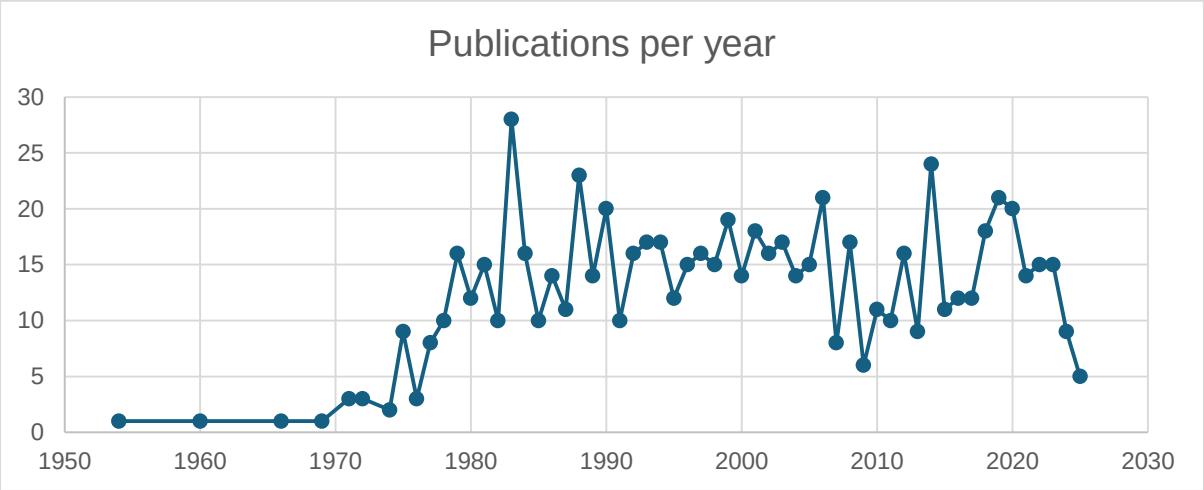

Fig 4. Publications per year. Highest publications in the year 1983 (23 papers), followed by 2014 (26), both 2019 and 2020 (25).

Highest citation was 2197 on human milk; 1646 citations on Chemical diversity in the sialic acids and related α-keto acids: an evolutionary perspective (2002); 1327 citations on Protein glycosylation: structural and functional aspects; 1014 citations on Selected indigestible oligosaccharides affect large bowel mass, cecal and fecal short-chain fatty acids, pH and microflora in rats (1997); 1101 citations on Bacterial metabolism and health-related effects of galacto-oligosaccharides and other prebiotics (2008); 907 citations on Matrix-assisted laser desorption/ionization mass spectrometry of carbohydrates (1999); 883 citations on Diversity in the sialic acids (1992); 652 citations on Advances in the biology and chemistry of sialic acids (2010). These citations indicated many research were done on oligosaccharidoses, however a few articles no data on citations.

4.**Discussions**

From our bibliometric analysis, most frequent themes were inborn error, gangliosidosis, carbohydrate, Pompe disease. All are related to oligosaccharidoses, included sialic acid storage disease (occurences 5). Some oligosaccharidoses were fatal, therefore required rapid diagnoses and treatment, e.g. enzyme replacement therapy. Some diseases were untreatable and patients remain very sick at home such as GM1-gangliosidosis patients. These disorders were classified into infantile-, juvenile- and adult-type with severe to mild symptoms.

Sewell, most active authors from our analyses, was the first scientist improved TLC method with processed with Dowex resin in 1981 (Sewell, 1981). Oligosaccharidoses detected included mucolipidoses I (sialidosis), Mannosidosis, fucosidosis, aspartylglucosaminuria, glycogen storage disease VI, β-galactosidase deficiency or GM1-gangliosidosis. Publication

included (9) 68 single authors with highest authorship of 17 authors. Single author may render not reliant while many authors may involve passive authors.

More than 150 publications were from United States, followed by Asian, European and Australian authors. Among Asian countries, Japan was most active in oligosaccharidoses publications, followed by China (nine publications), two publications by Malaysia (Rimshah, S et al., 2016; and Fatimah Diana et al., 2023), another two by our neighbouring countries, Singapore: Academy of Medicine, Singapore and Department of Pathology, Singapore General Hospital.
First oligosaccharidosis paper was found published in 1954 by by French Dexter, from United States, in Advances in carbohydrate chemistry, in google scholar search. This paper was on raffinose family of oligosaccharidosis. Raffinose was trisaccharides include glucose, galactose, fructose, an indicator for oligosaccharidoses, Rf Value.

There were about 40 oligosaccharidoses analysed included GM1-gangliosidosis (7 cases), GM2-gangliosidosis (2), Tay-sachs (2), fucosidosis (7), Pompe's Disease (9), Gaucher (4), Krabbe (1), Fabry disease (1), β-mannosidosis (1), I-cell (7) in Malaysia. This method was ardous and need expertise in interpretation (Peelen et al., 1994). Milk oligosaccharides were seen in some premature samples (Sewell, 2008).

We detected more cases year by year since 2007 until now. New methods were introduced such as enzyme assays, High Performance Liquid Chromatography (HPLC), Tandem Mass Spectrometry (TMS) for latent-onset patients and low samples volume. Molecular analysis was done for confirmatory of positive tests.

5. **Conclusions**

Bibliometric studies are important for analysis of the publication trends of oligosaccharidoses from world scientists. These analyses will help in improving diagnostic clue, methods, and treatment of oligosaccharidoses patients such as graft transplantation and enzyme replacement therapy. More financial supports were required for oligosaccharidoses research.

Acknowledgements
We would like to thank the Director General of Health Malaysia for his permission to publish this article.

Ethics approval
Exempted.

Conflict of interests
The authors declare that they have no conflict of interests.

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