

Two decades of echinococcosis/hydatidosis research: Bibliometric analysis based on the web of science core collection databases (2000–2019)

Mahdi Fakhar^{a,b}, Masoud Keighobadi^a, Hajar Ziaei Hezarjaribi^{a,*}, Mahbobeh Montazeri^{a,*}, Elham S. Banimostafavi^{a,b}, Shahram Sayyadi^c, Mohammad M. Ghaffari Hamadani^c, Ali Sharifpour^{a,b}, Rabeeh Tabaripour^a, Samira Asadi^a, Masoud Soosaraei^a, Ali A. Khasseh^d

^a Toxoplasmosis Research Center, Communicable Diseases Institute, Iranian National Registry Center for Lophomoniasis and Toxoplasmosis, Department of Parasitology, School of Medicine, Mazandaran University of Medical Sciences, Sari, Iran

^b Iranian National Registry Center for Hydatid Cyst (INRCHC), Department of Radiology, Imam Khomeini Hospital, Mazandaran Branch, Mazandaran University of Medical Sciences, Sari, Iran

^c Iranian National Registry Center for Hydatid Cyst (INRCHC), Department of Surgery, Mazandaran Branch, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran

^d Department of Knowledge and Information Sciences, Payame Noor University, Tehran, Iran

ARTICLE INFO

Keywords:

Bibliometric
Scientific collaboration
Echinococcosis
Hydatidosis
Echinococcus granulosus sensu lato
Echinococcus multilocularis
Web of science

ABSTRACT

Echinococcus granulosus sensu lato and *Echinococcus multilocularis* are responsible for serious health and economic implications for humans and animals. This study was designed to conduct a bibliometric analysis of global research on echinococcosis/hydatidosis included in the Web of Science Core Collection databases from 2000 to 2019. A total of 7066 relevant articles between 2000 and 2019 were identified. Most articles were published in 2015 (502 articles), 2017 (492 articles) and 2018 (493 articles), with the *Veterinary Parasitology* journal publishing the largest number of articles (237). Researchers from Xinjiang Medical University, China authored the most articles (388) in the field. Authors Craig, P.S. and Deplazes, P. were the most active in publishing 143 and 126 hydatid cyst research papers, respectively. The most echinococcosis/hydatidosis publications originated from Turkey, China and Iran, with 1210, 708 and 531 articles, respectively. The highest levels of research collaboration were evident between China-England, China-France, England-France, China-Australia, and China-Japan. Also, the top researchers in this field had relatively extensive collaborations with each other. Our bibliometric analysis provides a picture of the scientific research into the echinococcosis/hydatidosis field. Further multi-national collaborative research efforts in this field should show promising progress in the future.

* Corresponding authors at: Toxoplasmosis Research Center, Communicable Diseases Institute, Iranian National Registry Center for Lophomoniasis and Toxoplasmosis, School of Medicine, Mazandaran University of Medical Sciences, Farah-Abad Road, PO Box: 48471-91971, Sari, Iran.

E-mail addresses: ziaei2000@yahoo.com (H.Z. Hezarjaribi), m.montazeri2012@yahoo.com (M. Montazeri).

<https://doi.org/10.1016/j.fawpar.2021.e00137>

Received 29 May 2021; Received in revised form 31 October 2021; Accepted 7 November 2021

Available online 12 November 2021

This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Echinococcosis is a severe neglected zoonotic tapeworm disease caused by *Echinococcus granulosus sensu lato* and *Echinococcus multilocularis* producing cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively (Mc Manus et al., 2012). These parasites are responsible for serious health and economic implications in humans and animals (Deplazes et al., 2017). The World Health Organization (WHO) (2021) estimates that over 1 million individuals are affected by echinococcosis worldwide each year (World Health Organization (WHO), 2021), and Central Asia, Mediterranean countries, eastern Africa, South America, and western China are high endemic areas (Craig et al., 2007; Mahmoudi et al., 2019).

Humans are accidental intermediate hosts infected via fecal-oral routes by close contact with definitive hosts (domestic and wild canids) or by ingestion of the parasites' embryonated eggs in contaminated food, water, or soil, and fomites (Tamarozzi et al., 2020). Definitive hosts are infected through the ingestion of viable hydatid cysts in the infected viscera of intermediate hosts, which involves a wide range of mammals (Thompson, 2017). The liver (75%) and lungs (15%) are the most common locations for hydatid disease. However, the spleen, kidney, peritoneal cavity, the skin and muscles and rarely in the heart, vertebral column, brain, and ovaries have also been reported (Mc Manus and Zhang, 2003).

Imaging methods are essential for diagnosis, population screening and monitoring hydatid cysts in humans. Ultrasound and X-ray are widely used to diagnose CE or AE liver lesions and lung cysts, respectively (Bresson-Hadni et al., 2006; Solomon et al., 2018; Tamarozzi et al., 2018). Serodiagnostic assays as a confirmatory step with various levels of sensitivity/specificity can be helpful for detection of antigens or antibodies in definitive and intermediate hosts (Kern et al., 2017). The current recommendations for echinococcosis treatment include a combination of surgical and drug treatments with a long-term follow-up of cases (Brunetti et al., 2010; Salm et al., 2019).

The medical and economic importance of echinococcosis and the difficulty of diagnosis and treatment strategy is an important topic of investigation. Progress in control programs for diagnosis and development of effective drugs and vaccines is essential. Such progress will have a direct impact in the future on the control of echinococcosis, which continues to be a global challenge (Wen et al., 2019).

Bibliometric analysis is a scientific computer-assisted review methodology that can be an invaluable guide for a complete overview of a specific research field (Wen et al., 2019). Thus, the current study attempts to illustrate the global scientific publications on echinococcosis/hydatidosis in the web of science core collection databases from 2000 to 2019 using a scientometric approach.

2. Methods

The articles were retrieved from the web of science core collection databases, including SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, and IC. These databases are the most frequently used source of scientific information in the world (Ma et al., 2019). The document types (article, review and proceedings paper) published between 2000 and 2019 were included in this study. The keywords used included, "hydatid cyst*" OR "*Echinococcus granulosus*" OR "*Echinococcus multilocularis*" OR "echinococcosis" OR "Hydatidosis" OR "echinococcal disease". This search was performed in the topic field. If any of the above listed keywords were present in the title/abstract/keyword of the document, it was considered as one of the records related to echinococcosis/hydatidosis. The data were retrieved on June 1, 2020. According to the search strategy used in this study, a total of 7066 publications were obtained. Next, the bibliographic data were extracted and then analyzed using scientometric software.

The h-index is capable of simultaneously considering two important indexes, namely the number of articles and the number of citations; it can also evaluate the effectiveness of a researcher (Donthu et al., 2021). In this study, BibExcel software (Persson et al., 2009) was employed for H-index calculation and based on this, the top researchers were introduced. Also, the geographical distributions of research were determined based on the affiliation of the authors. Moreover, VOSviewer (Van Eck and Waltman, 2010) and the bibliometrix R Package (Aria and Cuccurullo, 2017) were used to assessment the international collaboration and co-authorship network of researchers in the hydatidosis field. Co-authorship is the most formal manifestation of intellectual collaboration between authors in the production of scientific research, which is the participation of two or more authors in the production of a work (Hudson, 1996). In this study, co-authorship means that two authors have published at least one joint article in the field of echinococcosis/hydatidosis.

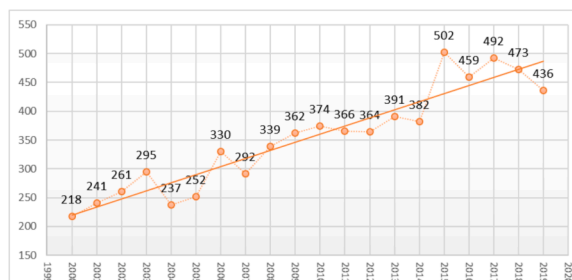


Fig. 1. Number of articles published on hydatidosis research annually between 2000 and 2019 in the web of science database. The highest number of articles were published in 2015, 2017 and 2018, respectively.

3. Results

From 2000 to 2019 (two decades), a total of 7066 articles in the field of hydatidosis have been indexed in the Web of Science database. As Fig. 1 shows, research in this area and over this period has been increasing. By year, the most articles were published in 2015 (502 articles), 2017 (492 articles) and 2018 (493 articles), respectively. The findings show that researchers from Xinjiang Medical University of China published the most articles (388) in the field, and the University of Zurich and the University of Bern in Switzerland ranked second and third with 258 and 243 articles, respectively.

3.1. Top journals in hydatidosis field

Between 2000 and 2019, the *Veterinary Parasitology* journal published the largest share with a total of 237 articles in the field of hydatidosis. Meanwhile, *Parasitology Research* and *Acta Tropica* journals ranked second and third with 233 and 167 papers, respectively. Also, journals such as *Parasitology Research* and *Iranian Journal of Parasitology* have increasingly focused on hydatidosis research (Table 1).

3.2. Top researchers with most published papers in hydatidosis field

Initial analysis of the data indicated that a total of 39,865 co-authors contributed to 7066 articles in the hydatidosis field. Also, 270 articles were single-authored and other articles had more than one author. Craig et al., 2007, with 143 papers, was the most active researcher in hydatid cyst research. Wen et al., 2019 ranked second with 136 articles, and Bruno Gottstein was third with 133 articles. Fig. 2 shows the names of the 15 leading hydatidosis researchers.

Among all authors of hydatidosis papers, Deplazes et al., 2017 was ranked first for H-index with a score of 40 (Table 2). This indicates that out of 126 articles published by this author, 40 papers received at least 40 citations.

3.3. Geographical distribution of research in hydatidosis field

The extent of each country's share in the research published on hydatidosis was another issue studied. Our results showed that researchers from Turkey published more articles (1210) compared to other countries, and accounted for more than 17% of the world's total hydatid cyst publications. China and Iran ranked second and third, with 708 and 531 articles, respectively (Fig. 3).

Fig. 4a and b illustrate the cooperation between countries. The thickness of the line between countries reflects greater cooperation. Data analysis showed that the researchers from China-England (92 cases), China-France (91 cases), England-France (79 cases), China-Australia (77 cases), and China-Japan (68 cases) had the highest level of collaboration.

Fig. 5 also shows the countries that received the highest citations in the field of hydatidosis research. Turkish authors received the most citations in this field, followed by authors from Switzerland and Australia.

3.4. Co-authorship network of researchers in hydatidosis field

Fig. 6 shows the co-authorship network of top researchers in the field of hydatidosis. In this study, the authors with more than 25 articles included in the network. The findings showed that the most co-authorship occurred between Ito A. and Sako Y. (44 cases) followed by Ito A. and Nakao M. (43 cases), Brunetti E. and Tamarozzi F. (32 cases), and Giraudoux P. and Raoul F. (27 items).

4. Discussion

In this study, we performed a comprehensive analysis of the worldwide trends in research outputs in the field of echinococcosis/hydatidosis from 2000 to 2019. Our analysis reveals an increasing publication trend in this period. In a previous study, it was reported that the average number of published articles from 2008 to 2017 was the highest among the periods from 1980 to 1996 and 1997 to 2007 (Ma et al., 2019).

Table 1

The top 10 journals in hydatidosis field. The three top journals published the largest number of articles.

Rank	Journal title	Articles published
1	Veterinary Parasitology	237
2	Parasitology Research	233
3	Acta Tropica	167
4	Parasitology	133
5	PLOS Neglected Tropical Diseases	130
6	Parasitology International	112
7	American Journal of Tropical Medicine and Hygiene	97
8	Iranian Journal of Parasitology	94
9	Experimental Parasitology	94
10	Journal of Helminthology	91

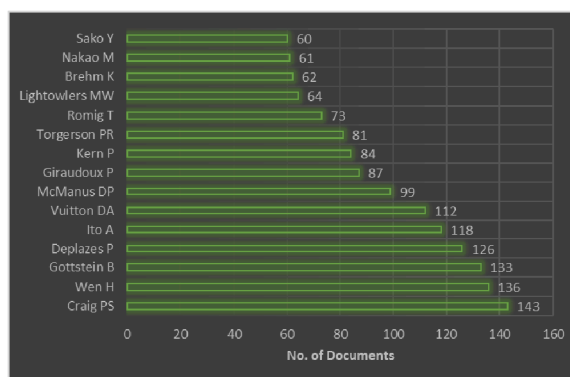


Fig. 2. The top 15 authors who contributed to most publications on hydatidosis.

Table 2

The top 20 hydatidosis investigators based on H-index.

H-index	Name	All citations	All articles
40	Peter Deplazes	6225	126
38	Philip S Craig	4447	143
38	Dominique Angèle Vuitton	4753	112
37	Akira Ito	3893	118
36	Bruno Gottstein	3457	133
34	Paul Torgerson	3582	81
33	Donald McManus	4053	99
31	Patrick Giraudoux	2496	87
30	Thomas Romig	2869	73
30	Minoru Nakao	2337	61
27	Petra Kern	3125	84
27	Klaus Brehm	2031	62
27	Yasuhito Sako	1793	60
26	Marshall Lightowlers	2043	64
25	Andrew Hemphill	1401	53
23	Renate Fink	1269	44
23	Andrew Thompson	2026	28
22	Hao Wen	1860	136
22	Solange Bresson-Hadni	1506	51
22	Peter M. Schantz	2138	31

The h-index is one of the most common criteria used to identify the impact of researchers in a field (Donthu et al., 2021). The present study is the first to show the cumulative impact of echinococcosis/hydatidosis research output by researchers. In this respect, Deplazes et al., 2017 with an h-index of 40 was considered to have the highest rank, followed by Craig PS. and Vuitton DA with an h-index of 38. Although the number of published papers by Craig PS. was higher than those published by Deplazes P., the papers published by Deplazes P. were more cited, and thus had a higher impact on related research. Interestingly, although Wen H. is the second most published researcher in the field of hydatidosis, the corresponding h-index is relatively low.

Our findings show that the top 5 countries in this field from 2000 to 2019 were Turkey, China, Iran, Germany, and the USA. Although, another report (Ma et al., 2019) that performed a bibliometric evaluation of echinococcosis research from 1980 to 2017, indicates that the top productive countries were Turkey, France and China. Also, based on a bibliometric analysis of hydatid cyst surgery literature from 1975 and 2017, Turkey, India and UK were found to be the most productive countries (Muslu and Senel, 2019). Similar to these previous studies, our data ranked Turkey first in the field of hydatid cyst publication numbers. Furthermore, the three most productive institutions were Xinjiang Medical University of China, the University of Zurich and the University of Bern. Another report (Ma et al., 2019), indicated that the top five institutions were the universities of Zurich (Germany), Bern (Switzerland), Salford (England), Franche Comte (France), and Xinjiang Med University (China). These data suggest that advanced levels and techniques of echinococcosis research might occur in these countries and institutions.

Although our findings show that Turkish researchers received the most citations, their overall average citation was not high. Compared to the studies conducted in other countries, Swiss researchers had the highest average citation score.

One of the most important issues in today's research is the collaboration between researchers, which eventually leads to the co-authoring of articles (Donthu et al., 2021). Contemporary research is moving towards increased collaboration. Our data indicated that many authors working on echinococcosis tended to collaborate with two or more authors regularly. It is evident that the top researchers from China-England, China-France, England-France, China-Australia and China-Japan had relatively large collaborations with each other in the field of echinococcosis/hydatidosis. However, it was previously suggested that there has been very little

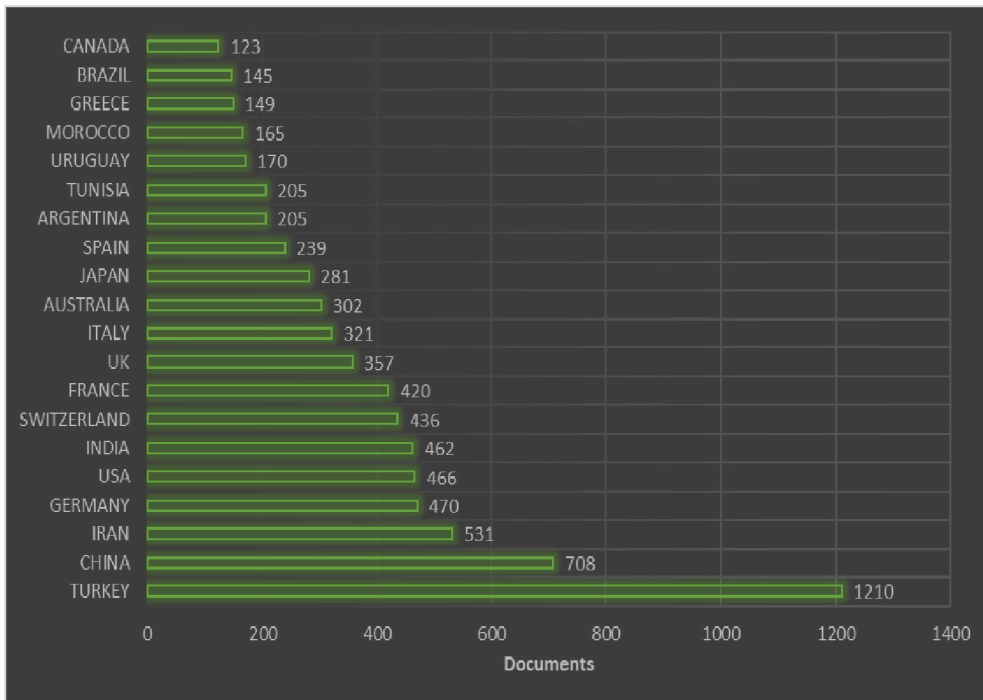


Fig. 3. The top 20 countries with the most publications on hydatidosis.

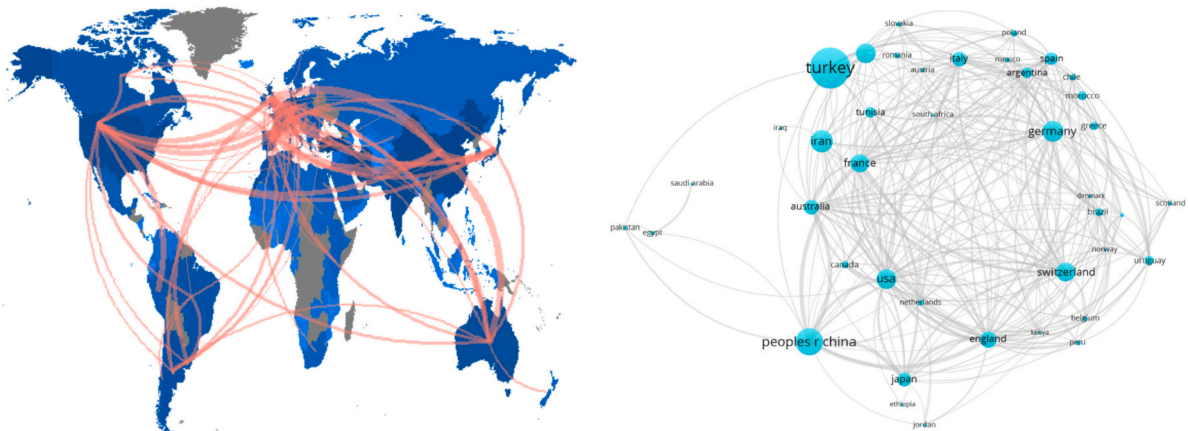


Fig. 4. a. International collaboration on hydatidosis as reflected in co-authorships. b. International collaboration on hydatidosis publications. The researchers from China-England, China-France, England-France, China-Australia, and China-Japan had the highest level of collaboration.

cooperation between the institutions in this field (Ma et al., 2019). The findings clearly indicate that the number of published articles on echinococcosis/hydatidosis is increasing and there is extensive scientific communication among researchers. There is promising potential for collaborating researchers from many different countries to make progress in different aspects of studies on echinococcosis/hydatidosis. On the other hand, it should be noted that although information regarding the geographical distribution of echinococcosis/hydatidosis could highlight potential hotspots for the diseases, the researchers' distribution might not necessarily match the actual location of the diseases. Thus, this should be evaluated separately in a future study.

Ethics approval

This study was approved by the research ethics committee of Mazandaran University of Medical Sciences (IR.MAZUMS.REC.1398.6300).

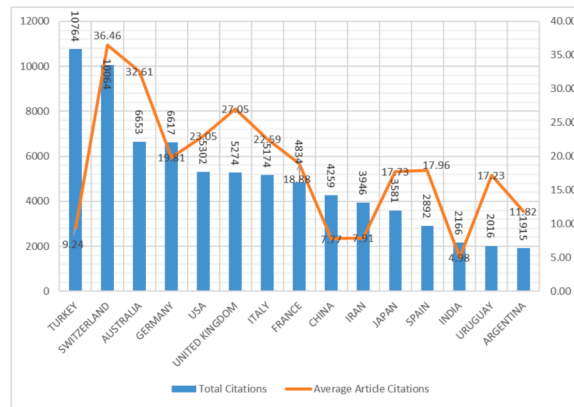


Fig. 5. Most cited hydatidosis articles according to countries of authors.

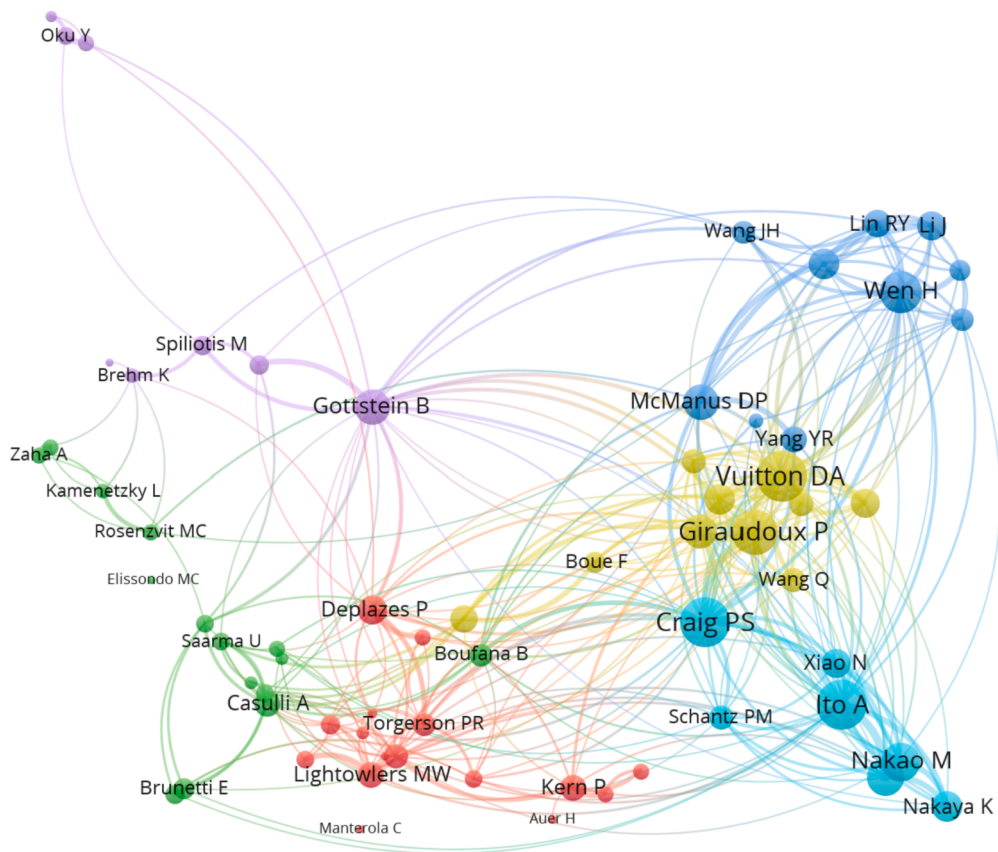


Fig. 6. Co-authorship network of researchers in hydatidosis field. The thickness of the lines linking authors indicates the amount of collaboration.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

This work was financially supported by the Toxoplasmosis Research Centre (TRC), Mazandaran University of Medical Sciences (Grant No. 6300).

References

- Aria, M., Cuccurullo, C., 2017. Bibliometrix: an R-tool for comprehensive science mapping analysis. *J. Inf. Secur.* 11 (4), 959–975. <https://doi.org/10.1016/j.joi.2017.08.007>.
- Bresson-Hadni, S., Delabrousse, E., Blagosklonov, O., Bartholomot, B., Koch, S., Miguet, J.-P., Manton, G.A., Vuitton, D.A., 2006. Imaging aspects and non-surgical interventional treatment in human alveolar echinococcosis. *Parasitol. Int.* 55, S267–S272. <https://doi.org/10.1016/j.parint.2005.11.053>.
- Brunetti, E., Kern, P., Vuitton, D.A., 2010. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop.* 114 (1), 1–16. <https://doi.org/10.1016/j.actatropica.2009.11.001>.
- Craig, P.S., McManus, D.P., Lightowlers, M.W., Chabalgoity, J.A., Garcia, H.H., Gavidia, C.M., Gilman, R.H., Gonzalez, A.E., Lorca, M., Naquira, C., 2007. Prevention and control of cystic echinococcosis. *Lancet Infect. Dis.* 7 (7), 385–394. [https://doi.org/10.1016/S1473-3099\(07\)70134-2](https://doi.org/10.1016/S1473-3099(07)70134-2).
- Deplazes, P., Rinaldi, L., Rojas, C.A., Torgerson, P., Harandi, M., Romig, T., Antolova, D., Schurer, J., Lahmar, S., Cringoli, G., 2017. Global distribution of alveolar and cystic echinococcosis. *Adv. Parasitol.* 95, 315–493. <https://doi.org/10.1016/bs.apar.2016.11.001>.
- Donthu, N., Kumar, S., Mukherjee, D., Pandey, N., Lim, W.M., 2021. How to conduct a bibliometric analysis: an overview and guidelines. *J. Bus. Res.* 133, 285–296. <https://doi.org/10.1016/j.jbusres.2021.04.070>.
- Hudson, J., 1996. Trends in multi-authored papers in economics. *J. Econ. Perspect.* 10 (3), 153–158. <https://doi.org/10.1257/jep.10.3.153>.
- Kern, P., Da Silva, A.M., Akhan, O., Müllhaupt, B., Vizcaychipi, K., Budke, C., Vuitton, D., 2017. The echinococcoses: diagnosis, clinical management and burden of disease. *Adv. Parasitol.* 96, 259–369. <https://doi.org/10.1016/bs.apar.2016.09.006>.
- Ma, X., Zhang, L., Wang, J., Luo, Y., 2019. Knowledge domain and emerging trends on echinococcosis research: a scientometric analysis. *Int. J. Environ. Res.* 16 (5), 842. <https://doi.org/10.3390/ijerph16050842>.
- Mahmoudi, S., Mamishi, S., Banar, M., Pourakbari, B., Keshavarz, H., 2019. Epidemiology of echinococcosis in Iran: a systematic review and meta-analysis. *BMC Infect. Dis.* 19 (1), 1–19. <https://doi.org/10.1186/s12879-019-4458-5>.
- Mc Manus, D., Zhang, W., Bartley, Li J.P., 2003. Echinococcosis. *Lancet.* 362, 1295–1304. [https://doi.org/10.1016/S0140-6736\(03\)14573-4](https://doi.org/10.1016/S0140-6736(03)14573-4).
- Mc Manus, D., Zhang, W., Bartley, Li J.P., 2012. Diagnosis, treatment, and management of echinococcosis. *Bmj* 344. <https://doi.org/10.1136/bmj.e3866>.
- Muslu, U., Senel, E., 2019. Bibliometric analysis of hydatid cyst and hydatid cyst surgery literature between 1975 and 2017. *Jentashapir J. Health Res.* 10 (2), e90859. <https://doi.org/10.5812/jjhr.90859>.
- Persson, O., Danell, R., Schneider, J.W., 2009. How to use Bibexcel for various types of bibliometric analysis. In: *Celebrating Scholarly Communication Studies: A Festschrift for Olle Persson at his 60th Birthday*, 5, pp. 9–24.
- Salm, L., Lachenmayer, A., Perrodin, S., Candinas, D., Beldi, G., 2019. Surgical treatment strategies for hepatic alveolar echinococcosis. *Food Waterborne Parasitol.* 15, e00050.
- Solomon, N., Zeyhle, E., Subramanian, K., Fields, P.J., Romig, T., Kern, P., Carter, J.Y., Wachira, J., Mengiste, A., MacPherson, C.N., 2018. Cystic echinococcosis in Turkana, Kenya: 30 years of imaging in an endemic region. *Acta Trop.* 178, 182–189. <https://doi.org/10.1016/j.actatropica.2017.11.006>.
- Tamarozzi, F., Akhan, O., Cretu, C.M., Vutova, K., Akinci, D., Chipeva, R., Ciftci, T., Constantin, C.M., Fabiani, M., Golemanov, B., 2018. Prevalence of abdominal cystic echinococcosis in rural Bulgaria, Romania, and Turkey: a cross-sectional, ultrasound-based, population study from the HERACLES project. *Lancet Infect. Dis.* 18 (7), 769–778. [https://doi.org/10.1016/s1473-3099\(18\)30221-4](https://doi.org/10.1016/s1473-3099(18)30221-4).
- Tamarozzi, F., Deplazes, P., Casulli, A., 2020. Reinventing the wheel of *Echinococcus granulosus* sensu lato transmission to humans. *Trends Parasitol.* 36 (5), 427–434. <https://doi.org/10.1016/j.pt.2020.02.004>.
- Thompson, R., 2017. Biology systematics of *Echinococcus*. *Adv. Parasitol.* 95, 65–109. <https://doi.org/10.1016/bs.apar.2016.07.001>.
- Van Eck, N.J., Waltman, L., 2010. Software survey: VOSviewer, a computer program for bibliometric mapping. *Scientometrics.* 84 (2), 523–538.
- Wen, H., Vuitton, L., Tuxun, T., Li, J., Vuitton, D.A., Zhang, W., McManus, D.P., 2019. Echinococcosis: advances in the 21st century. *Clin. Microbiol. Rev.* 32 (2), e00075-18. <https://doi.org/10.1128/CMR.00075-18>.
- World Health Organization (WHO), 2021. Characteristics and Details of Echinococcosis. <http://www.who.int/mediacentre/factsheets/fs377/en/>.