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Causality and Speech, Language and Hearing Sciences: epidemiological approach

Causalidade e Fonoaudiologia: abordagem epidemiológica

Keywords

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Descritores

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ABSTRACT

Purpose: To review the Speech-Language Pathology literature studies from the epidemiology and causality perspective. **Research strategies:** A national and international literature survey was carried out with searches from PubMed, SciELO and gray literature bases, conducted according to the instructions of the Cochrane Collaboration and published until January 9th, 2019. The review guiding question asks if Speech-Language Pathology uses methods in their evidence to infer causality. **Selection criteria:** All studies that presented a causal epidemiological approach in speech therapy were included, as well as excluded those that did not present an appropriate methodological approach for cause and effect analysis. **Data analysis:** Two authors of this study independently reviewed all citations. A priori determined form was used to extract the following data: author, year of publication, country of origin, theoretical conception, application or not of the study and central discussion addressed in the article. **Results:** From the search performed 3842 articles were found. However, none of them investigated their outcomes from the causality point of view, not allowing cause and effect inference. **Conclusion:** There is a shortage of studies that evidence causality in Speech-Language Pathology, which may alter the effectiveness and reliable handling of diagnosis and speech-language therapy, since it is still based on association and not on cause and effect based on studies designed to that.

RESUMO

Objetivo: Revisar a literatura dos estudos fonoaudiológicos sob a ótica da epidemiologia, segundo a perspectiva da causalidade. **Estratégia de pesquisa:** Realizou-se um levantamento nas literaturas nacional e internacional com buscas realizadas nas bases PubMed, SciELO e literatura cinzenta, conduzido segundo as instruções da Colaboração Cochrane e publicados até 8 de janeiro de 2019. A pergunta norteadora da revisão indaga se a Fonoaudiologia faz uso dos métodos epidemiológicos em suas evidências para inferir causalidade. **Critérios de Seleção:** Foram incluídos todos os trabalhos que apresentassem abordagem epidemiológica de causalidade em fonoaudiologia, assim como se excluíram os que não apresentassem abordagem metodológica adequada à análise de causa e efeito. **Análise dos dados:** Dois autores deste estudo, de maneira independente, revisaram todas as citações. Utilizou-se um formulário determinado a priori para extrair os seguintes dados: autor, ano de publicação, país de origem, concepção teórica, aplicação ou não do estudo e discussão central abordada no artigo. **Resultados:** Mediante a busca realizada, foram encontrados 3.842 artigos. Contudo, destes nenhum investigou seus desfechos a partir da ótica da causalidade, não permitindo a inferência de causa e efeito. **Conclusão:** Há escassez de estudos que evidenciem a causalidade na Fonoaudiologia, o que pode alterar a efetividade e o manuseio confiável do diagnóstico e a terapêutica fonoaudiológica, visto que ainda se baseia na associação e não na causa nem no efeito de delineamentos apropriados para tal.

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INTRODUCTION

Since long time ago, man has been concerned with unveiling the cause and effect relationship between the ordinary phenomena of life. Therefore, causality is the subject of study by epidemiologists and statisticians, conceptualized as a universal phenomenon that investigates the relationship between cause and effect in the same outcome if repeated⁽¹⁾.

Epidemiologically, causality is defined as the connection between the two processes, in which the first process is responsible for the occurrence of the second, or the presence of A causes B^(2,3), considering that every fact has a cause, and this cause produces the same effect under the same condition^(4,5).

The historical rescue of causality shows that during the man's understanding of the events occurring on earth, from myth to reason, philosophers such as Hippocrates explained the occurrence of diseases in the population with a rational foundation and exclusion of the supernatural in his treatise *Airs, Waters and Places*. Also, other theories were approached, explaining the influence of gods and nature, divine causes, among others⁽⁶⁾.

In the modern era, the evolution and organization of epidemiological thinking had several protagonists such as Hume, Mills, Locke, and Suppes, who contributed to the conception of causal inference⁽⁶⁾. Scholar John Snow had a recognized contribution to epidemiology through the Cholera Outbreak Essay, relating living conditions and disease processes. For him, to obtain an effective intervention to prevent the disease, it was necessary to know the distribution of cases in space and time (of occurrence), as well as the characteristics of the patients^(7,8).

According to this understanding, the association between epidemiological studies and economic, cultural, demographic, and social factors, also called as health determinants are the government's responsibility, and the therapeutic practices should be collective. Another relevant aspect in the evolution of understanding and the application of knowledge of causality as conceived in this century are the concepts of uni-causality and multi-causality, which play an important role in determining and combating diseases and complications⁽⁹⁾. According to the understanding that not all phenomena obey deterministic laws, and most of them are the product of multiple causes, it was possible to calculate the probability of occurrence of an event due to the presence or performance of one or several causes. Thus, probabilistic or stochastic models were built, able to know the risk factors and the advance in the elaboration of strategies in the health-disease process^(10,11).

Scientific knowledge is essential, aimed at providing information, enabling the analysis of the health-disease process, and defining more accurate preventive and therapeutic measures, resulting in improved clinical and collective practices⁽¹²⁻¹⁴⁾. Thus, the social aspects under the view of speech pathology need to understand the health-disease process in the population and not only in the individual, multiplying the production of knowledge.

Therefore, the objective of this paper is to systematically review the literature of speech therapy studies from the perspective of epidemiology, and the perspective of causality.

RESEARCH STRATEGY

This systematic literature review was performed according to the instructions of the Cochrane Collaboration.

The research question that supports this review was: Does speech therapy use epidemiological methods in its evidence to explain causality?

Selection criteria

Eligibility criteria

Only articles with the epidemiological approach of causality in speech therapy were included in the references found. Design of cohort observational studies, prospective and retrospective case-control studies without any language restrictions and published until January 9th, 2019 were accepted.

Exclusion criteria

Studies without an adequate methodological approach for cause and effect analysis were excluded.

Search strategy

The searches were performed in the electronic databases MEDLINE (accessed via PubMed), Scielo, and Google Scholar (gray literature). The search strategy used keywords identified in MeSH (Medical Subject Headings), DeCS (Descriptors in Health Sciences), as shown in Chart 1.

Study selection

Two authors of this study (RSR and NCS) independently reviewed all citations, abstracts, and articles found in the search to determine eligibility for study inclusion, blinded to each other's results. When the reviewers were unable to determine the inclusion/exclusion on the abstracts, the article was accepted for full reading to determine its possible inclusion. Disagreements at all stages of the selection process will be resolved by an independent and blind third-party reviewer (BNGG).

Data analysis

Data extraction

A form was chosen to extract the following data: author, year of publication, country of origin, theoretical conception, application or not of the study, and central discussion addressed in the article.

RESULTS

As shown in the flowchart (Figure 1), the search strategy had 3,842 studies. None of them used a methodology for adequate cause and effect analysis.

The studies found had different topics, such as: the improved speech perception in noise with a current focus on cochlear implant users; the treatment of dysphonia in the elderly

population: the role of the speech therapist; the diagnosis and the treatment of post-extubation dysphagia: results of a national research; a causal link between visual spatial attention and reading acquisition; a practical guide for cochlear implantation in adults with long periods of monaural sound deprivation and screening for speech and language delay in preschoolers. None of the studies allowed a cause and effect analysis.

The predominance of scientific production was characterized by the theme in the area of children's language, with emphasis

on reading and writing, and the area of audiology. Most methods used were cross-sectional studies, literature reviews, and case studies. Moreover, the most used statistical analyses were descriptive, correlations and bivariate associations.

Since no approaches were found with the perspective of causality intended to analyze in this study, it was decided to perform a description of some studies on causality, aiming at the expansion of knowledge on the topic by the speech therapists (Chart 2).

Chart 1. A search strategy used on Medline accessed via PubMed, SciELO and Google Scholar (2019)

Index	Strategy	Hits
PubMed		
#1	"causality" [Mesh] OR (causal* AND (multifactorial))	763.826
#2	"speech" [Mesh] OR (speech AND (therapy OR disorder OR pathology)) OR "audiology" [Mesh] OR "hearing sciences" [Mesh] OR "hearing impairment" [Mesh] OR "deafness" [Mesh]	104.057
#3	# 1 AND #2	1.6142
#4	NOT cardiovascular) NOT gen*) NOT syndrome*) NOT molecular*) NOT animal) NOT brai*) NOT hybr*) NOT biomechanic*) NOT diseases*	-
#5 Search	# 3 AND #4	812
Complete search	(((((("causality"[Mesh] OR (causal* AND (multifactorial)))))) AND ((("speech"[Mesh] OR (speech AND (therapy OR disorder OR pathology)) OR "audiology"[Mesh] OR "hearing sciences" [Mesh] OR "hearing impairment" [Mesh] OR "deafness" [Mesh]))) NOT (cardiovascular) NOT gen*) NOT syndrome*) NOT molecular* NOT animal) NOT brai*) NOT hybr*) NOT biomechanic*) NOT diseases*)	812
SciELO		
Complete search	#(causalidade e fonoaudiologia) #(causalidade, fonoaudiologia e epidemiologia) #(fonoaudiologia, epidemiologia e saúde pública)	300
Scholar Google		
Complete search	(causalidade e fonoaudiologia) e (causalidade, fonoaudiologia e epidemiologia) e (fonoaudiologia, epidemiologia e saúde pública)	2.730

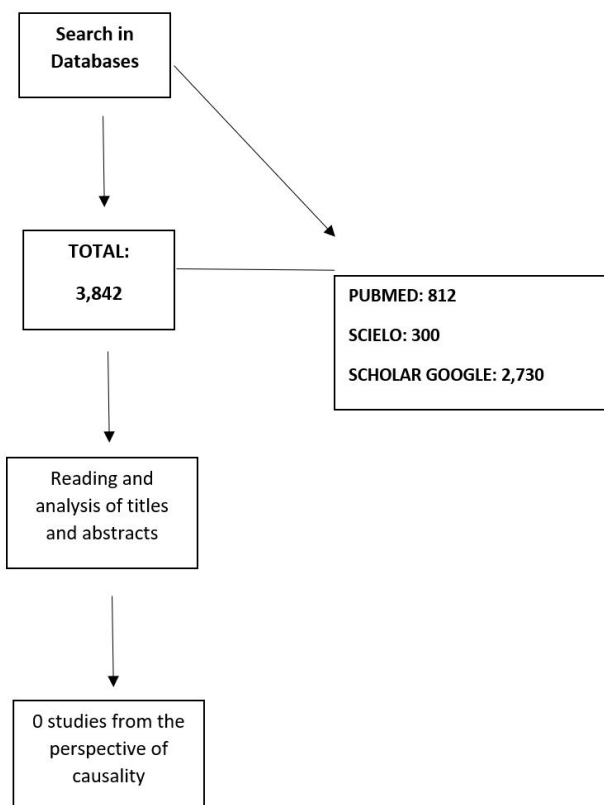


Figure 1. Flowchart of the electronic search of the analyzed studies, 2019

Chart 2. Characteristics of the studies selected for the systematic review accessed via PubMed, SciELO and Google Scholar (2019)

Authors (year, country)	Objective	Theoretical conception	Applied study	Main discussion
Mota and Kuchenbecker (2017) ⁽¹⁵⁾ ; Brasil	To demonstrate causal relationships between medications and adverse events, emphasizing the importance of Rothman's causality model.	Epistemological	Descriptive	Rothman model
Hamra et al. (2015) ⁽¹⁶⁾ ; USA	To verify the fitting sets with averaging technique on models obtaining causal estimates based on multiple models.	Statistical	Descriptive	ADG theory and multiple model calculation
Araújo et al. (2014) ⁽¹⁷⁾ ; Brasil	To review the probabilistic determinism, multi-causality from Hill and Mackie's perspective on causation.	Epistemological	Descriptive	Deterministic and probabilistic view of causality
Lagnado et al. ⁽¹⁸⁾ (2013); Inglaterra	To emphasize the interrelations between causality, contractual, and attributions of responsibility, proposing a new model of judgment.	Behavioral analytical	Descriptive	Causality and counterfactual theory
Marshall and Galea (2015) ⁽¹⁹⁾ ; USA	To elucidate agent-based models that can be used to simulate contractual output in the presence of complexity to assess causal effects.	Epistemological	Descriptive	Counterfactual causality inference
Petersen and van der Laan (2014) ⁽²⁰⁾ ; USA	To elucidate the usefulness of causal thinking, providing an accessible introduction to the flexible and powerful tools of the causal models.	Epistemological	Descriptive	Causality model
Tuner et al. (2013) ⁽²¹⁾ ; USA	Explain the conditions necessary to obtain signs of mental contracture based on the Rubin Model.	Epistemological	Descriptive	Rubin Causality Model and Causality Inference
Wang et al. (2013) ⁽²²⁾ ; USA	To explain the correlations between multiple mediators using continuous latent variables for any mediators through a joint distribution model.	Epistemological	Descriptive	Causality mechanism of multiple mediators
Plümper et al. (2010) ⁽²³⁾ ; Inglaterra	To show the causal inference of qualitative research using Monte Carlo techniques to evaluate the performance of different techniques or case selection algorithms with larger samples.	Statistical	Applied qualitative analysis	Monte Carlo technique and causality inference
Petersen et al. (2006) ⁽²⁴⁾ ; USA	To verify the estimation of direct natural effects by implementing statistical software.	Epistemological	Descriptive	Causality mechanism and statistical software
Rothman e Greenland (2005) ⁽²⁵⁾ ; USA	To clarify the causality model and the principles of multi-causality for sufficient causes and components.	Epistemological	Descriptive	Causality Model and Principles of Multi-causality
Hernán et al. (2004) ⁽²⁶⁾ ; USA	To show how the structure of causal diagrams in statistics leads to adjusting selection biases and helps to differentiate them.	Statistical	Descriptive	ADGs Theory
Olsen (2003) ⁽²⁷⁾ ; Dinamarca	To discuss ways in which the cause is seen in epidemiology and evaluate the concept of causality.	Epistemological	Descriptive	Epidemiological view on causality
Parascandola and Weed (2001) ⁽²⁾ ; USA	To review the epidemiological literature, seeking dominant patterns and definitions of chance.	Epistemological	Descriptive	Causality Theory
Susser (2000) ⁽²⁸⁾ ; USA	To review multi-pronged approaches on the causality theory.	Epistemological	Descriptive	Multi-causality Theory
Greenland (2000) ⁽²⁹⁾ ; USA	To review the counterfactual approach to causality analysis in health sciences, connections to graphical and structural equation approaches, and longitudinal data analysis.	Statistical	Descriptive	Counterfactual analysis of causality
Czeresnia et al. (1995) ⁽³⁰⁾ ; Brasil	To discuss the construction basis of the concept of risk by presenting Rubin's causality inference model developed within the scope of applied statistics.	Statistical	Descriptive	Rubin Model

Chart 2. Continued...

Authors (year, country)	Objective	Theoretical conception	Applied study	Main discussion
Cox (1992) ⁽³¹⁾ ; Inglaterra	To review the latest statistical thinking on the topic from the point of view that there is certainly some sense in which causality is central to the scientist's efforts to understand the real world.	Statistical	Descriptive	A statistical view of causality theory
Holland (1986) ⁽³⁾ ; USA	To investigate the contributions that the statistical model can make about causality.	Statistical	Descriptive	Statistical model
Stehbens (1985) ⁽³²⁾ ; Nova Zelândia	To discuss the classification and determination of the roles of associated factors involved in the development of a disease and multi-causality.	Epistemological	Descriptive	Causality Theory
Rothman (1976) ⁽³³⁾ ; USA	To show a schema for interrelating causes that can provide a useful way of thinking about effect modification as a description of nature.	Epistemological	Descriptive	Causality theory and causality model
Hill (1965) ⁽³⁴⁾ ; USA	To analyze whether environment and disease aspects are causal or associative based on Hill's criteria.	Epistemological	Descriptive	Causality Hill Criteria

DISCUSSION

The initial objective of this study was to review the literature systematically regarding speech therapy studies about causality. However, no articles were identified with this perspective, showing the weakness of the causal approach in specific studies of speech therapy. Thus, the description of some studies on causality aiming to expand the knowledge on the topic and discussed by the speech therapists was chosen.

The idea of a cause is not just predicting events but explaining them. It is possible to predict events based on correlations; however, A must explain B to assign causation. It is very difficult to know if and when A causes B⁽³⁵⁾. The development of mathematics and statistics has facilitated the process of causality. However, the simple observation of the occurrence of association or correlation between A and B cannot determine causality^(36,37). Only by observing that two phenomena are associated does not mean that one is causing the other, because various non-causal or spurious situations may occur^(19,25,38). Thus, speech therapy is still based on the association and not on the cause or proven effect, which can change the diagnosis and treatment.

The occurrence of spurious associations has deceived men throughout history, and until today, science struggles to identify causal mechanisms⁽³⁹⁾. Therefore, efforts by the speech-language scientific community are needed to improve the methods and analysis used.

Epidemiology has used both experimental and observational studies to identify causal factors⁽³⁵⁾. Regardless of the observational or experimental approach the researcher uses, a causal hypothesis should be incorporated *a priori* from a risk perspective and stipulating what criteria will be used to judge whether an association is causal or not, minimizing errors such as chance, biases, and confounders⁽²⁵⁾. The type of design used is another key piece to be considered as each method has its assumptions and the causality supply is different⁽¹⁰⁾.

The data described in Chart 1 show that most studies are published by the United States (56%), followed by England (17%), Brazil (13%), Italy, Denmark, and New Zealand (4%). The selected articles verified the definition of the concept of causality, the presentation of causal models and the relevance of statistics, although there may be some divergences in the theoretical conception of the authors. Also, according to the researched literature, the main approaches are of the theory of multi-causality, mostly with epistemological theoretical conception.

In the work of Rothman and Greenland⁽²⁵⁾, no cause is self-sufficient to trigger the disease. Thus, the theory of multi-causality can identify other components of a complex causal model, such as underlying disease, genetic predisposition, nutritional status, and age of the individual, involving several causal mechanisms.

The use of medications has been researched as an interference factor in the dynamics of adverse events. Mota and Kuchenbecker investigated whether drug use is associated with adverse effects by applying the Rothman Causality Model. In this model, the causal agent may be composed of a constellation of causes, sufficient of an adverse event. The model further explains that different causal components act in groups to produce an effect, necessarily implying that the component causes must act at the same time⁽¹⁵⁾.

The “complex causal networks” indicate that numerous elements played a role in producing a given effect or “causal web”, which, according to their elaboration, epidemiologists should increase their ability to describe and to study the complex interrelationships between risk factors and diseases, that is, the existing connection between all elements of the web directly or indirectly, however, they are all part of the same set⁽⁴⁰⁾.

The association does not imply a causality relationship, although there are several historical attempts to equalize the two concepts⁽¹⁷⁾. The probabilistic deterministic view of causality is the

understanding that statistics may or may not offer, connected to concepts such as strength, consistency, plausibility and specificity, approximation with analytical philosophy, and contributing to a better understanding of causal relationships⁽¹⁷⁾, that is, to identify that A causes B, A must exist before B, and for B to occur, A must be present before B; and in the absence of A, B does not occur, and there is some plausibility within existing knowledge that explains that A causes B. An association indicates that A is associated with B, but there is no cause determination or indication if A or B happened before.

There are some techniques to show causal inference, which are explored below: The Monte Carlo technique (MCT) is used to demonstrate the causal inference of qualitative research by assessing the performance of different techniques or case selection algorithms with larger samples. The basis of MCT is to generate values for the variables of the model to be studied by random numbers⁽⁴¹⁾.

The acyclic directed graph theory (ADG) shows how the structure of causal diagrams in statistics leads to adjusting selection biases and helps to differentiate them. The use of ADG in causal modeling confirms the idea that causality implies directionality of influence. The causal relationship is asymmetrical, in which the cause influences the outcome and never the opposite. In an ADG, the existence of an arrow between both variables A and B expresses the possibility of a direct causal relationship between them. Thus, the absence of an arrow between two variables A and B shows the most definitive hypothesis of null direct causal effect^(16,42).

For a good understanding of the determinants and conditioning factors in the health-disease process and its implications, a more detailed view at individual and collective living conditions is needed, including basic subsistence materials related to nutrition, housing, and sanitation⁽³⁹⁾, since such characteristics exert interaction, which can make certain individuals more vulnerable than others, influencing the course of an outcome, and that causality factors can be organized in a hierarchical, most proximal order to the distal socioeconomic factor⁽¹⁰⁾.

A study conducted in Norway addressed a population with caries and applied the theory of causality to verify the processes and justify their occurrence. The study showed that the lack of a specific model does not allow any results to be accepted for risk identification purposes and there is only a fragmented knowledge of the associations between social, contextual, individual and the caries variables⁽⁴³⁾.

The recognition of social determinants in the health-disease process by social epidemiology explains the status of a theory that facilitates the understanding of the systems that form the needs of social individuals, reorienting the mechanisms for epidemiological analysis, indication of diagnoses and the definition of preventive and therapeutic interventions⁽¹³⁾. This enhances the improvement of speech therapy practice in health, in fulfilling the role of taking care of people effectively, highlighting the individual and collective quality of life⁽³⁹⁾.

The speech therapy should proceed based on this, acting directly in research, seeking causal confirmation, and not only embracing the association as the veracity of occurrences. The area has all levels of health care and aims at the construction

of universality and comprehensiveness in its preventive and rehabilitative practices, individual or collective. However, the theoretical reference subsidizing its actions has limitations, such as not analyzing the health-disease process in its complexity, without encompassing the peculiarities of human communication (hearing, written/oral language, voice, fluency) and dysphagia. Evidence tends to neglect the particularities of the subjective, social, historical, and cultural processes of each context/community.

Therefore, from the perspective of the theory of causation, the speech therapist will have an analysis of the disease's thread and its progression more clearly, following along the path to the outcome and recognizing the extent of the effect. Thus, prevention and promotion actions will be handled reliably and effectively.

CONCLUSION

Studies in the field of speech therapy do not use the theory of causality, nor methodologies that explore it. However, based on the conceptual proposal described in this article, speech therapists will have substantial content to advance as a science.

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Authors contributions

NCS and RSR participated in the study design, data collection, analysis and interpretation, writing, and final review of the article. BNGG participated as an advisor, in the conception of the study, analysis, data interpretation, writing and final review of the article.