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Oropharyngeal Dysphagia: A Proposal for an Ecological Theoretical Model

A Theoretical and Conceptual Model for Oropharyngeal Dysphagia

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Design: This integrative literature review.

Setting: This was carried out until January 4, 2021, and searches were performed with the MEDLINE, EMBASE, and other bases.

Methods: The directed acyclic graphs were constructed from data retrieved from the selected literature.

Results: Among the 91 articles found that sought to identify theoretical models associated with oropharyngeal dysphagia, only eight articles have a theoretical model. Of these, only five presented an outcome directly related to swallowing, revealing potential confounding factors but no potential effect mediators. No study showed a directed acyclic graph. Thus, two directed acyclic graphs will be presented, one with the main associated factors that increase the probability of developing dysphagia and the other with related to the damaging results of changes in the safety and efficiency of the swallowing mechanism.

Conclusion: Dysphagia is complex and surpasses the understanding of the health-disease process at the individual level. It is known that population health is a product of ecological circumstances resulting from the interaction between human societies and the environment in general, as well as with their specific ecosystems and other support processes.

Keywords: deglutition, deglutition disorders, theoretical models, epidemiology.

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I. INTRODUCTION

Swallowing is a complex, semi-automatic, continuous, neurophysiological mechanism. It is mediated by brainstem nuclei that play a vital role in this process [1-3]. Dysphagia is a difficulty in swallowing [4,5].

Dysphagia can result from different etiologies, including neurological diseases, such as dementia [6], Parkinson's disease [7], multiple sclerosis [8], stroke [9], anatomical and physiological changes or other comorbidities, such as head and neck cancer [10], cervical spine surgery [11], traumatic brain injury [12] and chronic obstructive pulmonary disease [13]. Normal aging also presents a propensity to favor this symptom [14].

Dysphagia is a potential indicator of population health because, in addition to being one of the symptoms of several prevalent diseases in the elderly population, it can also manifest in clinical conditions that lead to hospitalization and hospital readmissions, such as dehydration, malnutrition, and aspiration pneumonia [15-17], as well as early mortality. It has also been associated with changes in social and emotional aspects since, in many Eastern and Western cultural contexts, mealtimes are usually an opportunity for human and social interaction and pleasure. To some degree, swallowing issues may interfere with the quality of life [18-21].

There are still no established estimates regarding the prevalence of dysphagia among the older people in the world literature and the inclusion of chronic and neurological diseases has contributed to the variability of these data, as well as heterogeneous diagnostic criteria and screening instruments of low methodological quality [19,22,23]. In some specific clinical studies, the prevalence of dysphagia has been reported to range between 8.1-80% among stroke patients and 11-81% among Parkinson's disease patients, and it appears in 27-30% of traumatic brain injury cases and 91.7% of patients with community-acquired pneumonia [24].

Therefore, given the importance of swallowing for human existence, the consequences of this condition, the direct or indirect health problems resulting from changes in swallowing, and the gap in the indexed

literature concerning theoretical models of oropharyngeal dysphagia, this article aims to propose a multi-causal model using directed acyclic graphs with mediating variables, confounding variables and variables with direct causation to oropharyngeal dysphagia.

II. METHODS

We performed an integrative literature review by searching the MEDLINE (accessed via PubMed), EMBASE, Scielo and Google Scholar electronic databases. Search strategies were adapted to each database, using MeSH (Medical Subject Headings), DeCS (Health Sciences Descriptors) and Emtree (Embase Subject headings) keywords related to the outcome. Some main keywords were: (((conceptual [All Fields] AND framework [All Fields]) OR ("models, organizational" [MeSH Terms] OR ("models" [All Fields] AND "organizational" [All Fields]) OR "organizational models" [All Fields] OR ("models" [All Fields] AND "organizational" [All Fields]) OR "models, organizational" [All Fields])) OR (theoretical [All Fields] AND ("Model Driven Eng Lang Syst" [Journal] OR "models" [All Fields]))) AND ("deglutition disorders" [MeSH Terms] OR ("deglutition" [All Fields] AND "disorders" [All Fields]) OR "deglutition disorders" [All Fields] OR "dysphagia" [All Fields]). In addition, we used the following keyword search to construct the directed acyclic graphs: "Deglutition Disorders" [Mesh] OR ((swallow * OR

deglutition OR Oropharyngeal) AND (problem * OR disorder * OR impairment * OR difficult * OR dysphagia) OR dysphagia). Both searches were carried out until January 4, 2021, with no language restrictions.

The directed acyclic graph was constructed on the <http://www.dagitty.net/dags.html> website, based on a careful analysis of the data in the indexed literature, and a theoretical causal model. This website offers free access online and offline via download. The online version of our directed acyclic graph was constructed by adding exposure and outcome variables, covariates, an ancestor of the exposure and an ancestor of the outcome for the theoretical model. From the arrows (causal relationships), we identified the variables to be adjusted, whether causal path or biased causal path variables.

III. RESULTS

Among the 91 articles in the review that sought to identify theoretical models associated with oropharyngeal dysphagia, only eight articles [33-41] presented a theoretical model. Of these, only five had outcomes directly related to swallowing and led to potential confounding factors. However, none showed potential effect mediators [33, 36-39] (Table 1). Among the 91 articles, only one had a longitudinal design [39], two were qualitative, 27 were integrative literature reviews, and the rest were cross-sectional studies. No study presented a directed acyclic graph.

Table 1: Characteristics of the studies found with theoretical models in the indexed literature. 2020.

Title	Study design	Outcome	Model
Oral hypofunction in the older population: Position paper of the Japanese Society of Gerodontology in 2016 [33]	Conceptual review	Oral hypofunction in an older population	Three large blocks, with the activities of the first one centered on the community and oral frailty. The middle block concerned the dentist and issues of oral hypofunction. The last block addressed oral disorders and, when necessary, the intervention of specialists to treat chewing and swallowing problems.
The consumption of snacks and soft drinks between meals may contribute to the development and persistence of gastro-esophageal reflux disease [34]	Conceptual review	Gastric acid secretion	Subsequent phases were related to consequences following the consumption of specific diets and quantities, resulting in exposure to acid secretion in the esophagus.
The Experience of Head and Neck Cancer Survivorship (Including Laryngectomy): An Integrated	Conceptual Review	Head and neck cancer	A pyramid with the disease and treatment at the base, building upward toward the main variable at the peak: quality of life. The second most important peak

Biopsychosocial Model [35]			variables were psychosocial factors.
Defining the End-Point of Mastication: A Conceptual Model [36]	Conceptual review	The end-point of mastication	Decision-making structure to define when it is safest to swallow food after chewing.
Understanding the Dining Experience of Individuals With Dysphagia Living in Care Facilities: A Grounded Theory Analysis [37]	Qualitative	Successfully feeding with dysphagia	An ecological social model to make mealtimes successful (dinner) for individuals with dysphagia, with individual, interpersonal and organizational variables.
Malnutrition and Clinical Outcome of 234 Head and Neck Cancer Patients Who Underwent Percutaneous Endoscopic Gastrostomy [38]	Cross-sectional	The survival of head and neck cancer patients who underwent percutaneous gastrostomy	Predictive anthropometric/laboratory parameters to differentiate between patients that survive endoscopic gastrostomy for <4 months and long-term survivors.
Adherence to Dysphagia Treatment Recommendations: A Conceptual Model [39]	Conceptual review	Adherence to dysphagia treatment	Three blocks with health factors, individual patient factors, and contextual factors (community, support technology), and a link between the interference of barriers or facilitating elements in the adherence to dysphagia treatment.
Swallowing Impairment in Older Adults: Association With Sensorimotor Peripheral Nerve Function From the Health, Aging and Body Composition Study [40]	Longitudinal	Swallowing impairment	A five-block hierarchy. The first concerns demographic and socioeconomic characteristics. The second, behavioral variables. The third, oral health. The fourth, general state of health. The fifth, impaired peripheral sensory and motor functions, with both arranged in a hierarchy of swallowing problems.

When analyzing the articles on oropharyngeal dysphagia, we found 77,918 results regarding different populations (pediatric, geriatric, neurological, institutionalized, genetic, and adult), with different themes, diagnostic and screening methods, rehabilitation plans, and designs. Regarding the factors we listed as the main ones for our research, we found six large groups associated with oropharyngeal dysphagia, namely: social, economic, demographic, behavioral, general health, and oral health factors. They will be presented next.

The main associated factors we found in the indexed literature can be divided into two large groups, those that increase the likelihood of dysphagia and those related to the damage caused by changes in the safety and efficacy of the swallowing process. This article will present a graph of the factors that increase the likelihood of developing oropharyngeal dysphagia.

The factors that are associated with a higher chance or prevalence of dysphagia demonstrate that exposure factors are individual characteristics (e.g., sex and ethnicity) [41], biological and physiological changes related to aging (loss of muscle mass and function, decreased tissue elasticity, sensory impairment and reduced compensatory capacity in the brain[42-44], health conditions (several chronic diseases, neurological diseases, deficiencies and use of medication) [45-48], oral health conditions (mainly tooth loss and xerostomia) [49-52], and socioeconomic issues (income, social vulnerability, health services) [53-55].

Decreased chewing efficiency (due to deficiencies in the dental arch or ill-fitting prostheses), xerostomia (self-perceived or due to underproduction), senile denervation of the esophagus, conditions that compromise visceral motor activity (due to neuropathies, myopathies, diabetes, etc.), and the use

of drugs which can compromise the muscle activity of the organs involved in swallowing, are also important factors that often contribute to the occurrence of dysphagia [56-58].

The directed acyclic graph (DAG) (Figure 1) was based on the studied literature, and the authors of this article thoroughly discussed it. In our analysis, we observed individual variables directly related to dysphagia [41, 42, 59-61], as well variables related to

health conditions such as neurological diseases, disability, and oral health problems [56-58]. Daily habits and health behaviors were indirectly [62-64], and directly linked to health conditions (chronic and neurological diseases), which are mediators or are directly associated with dysphagia [65-69]. Socioeconomic development, income, and social vulnerability are variables that precede health conditions, which are directly related to swallowing problems [53-55].

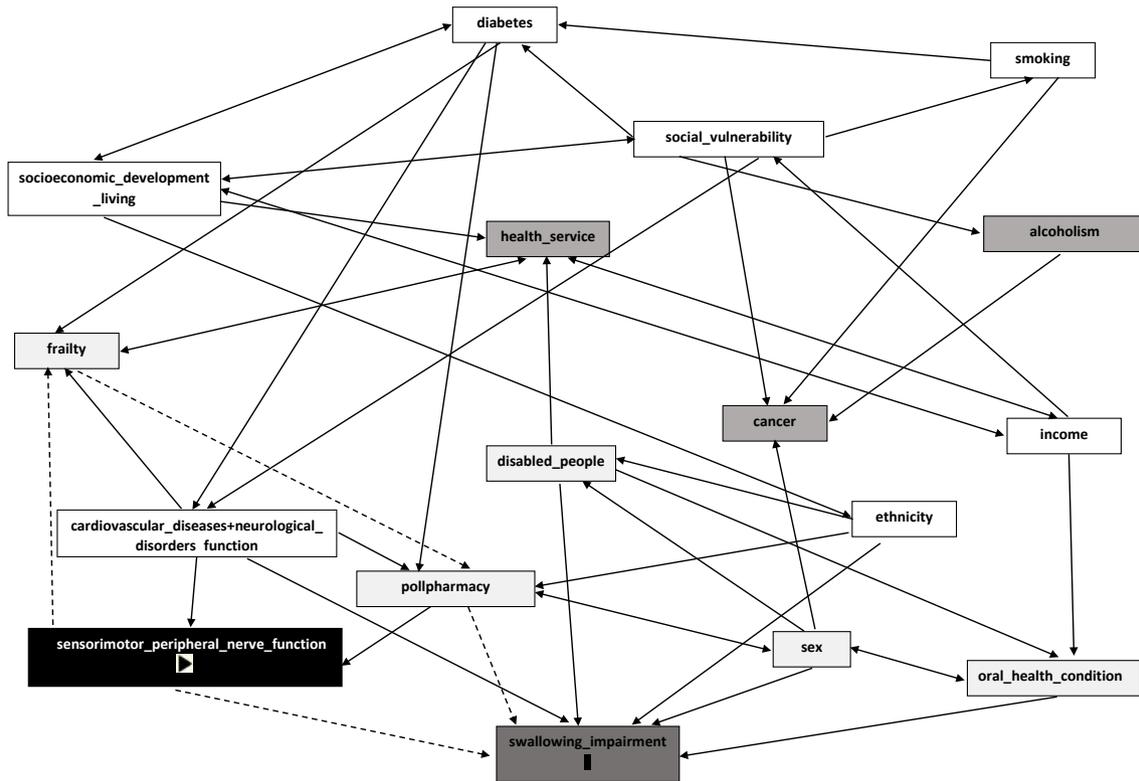


Figure 1: Directed acyclic graph (DAG) to show associations from the literature review.

Legend: Black Background ►: variable exposure; White background: variable adjusted; Dark gray background ■: variable outcome; Light gray background: variable ancestor of outcome; Medium gray background: another variable

In Figure 1, we observe that in multivariable analyses to control confounding factors (i.e. confounding bias), it is essential to control variables such as diabetes, cardiovascular and neurological disorders, socioeconomic development living, social vulnerability, smoking, ethnicity, and income. When these variables are not controlled, bivariate analyses can lead to erroneous conclusions regarding possible factors associated with non-true significance. After studying all the variables, we observed that changes in the peripheral nervous system constitute the only independent, directly associated variable, even though it may also be associated with stroke and neurological diseases. All others are influenced by variables that precede exposure and outcome. It is important to remember that it is mandatory that the ecological theoretical model takes into account the age of the population to be studied.

As for the factors related to the damage resulting from changes in the safety and efficacy of the swallowing process (Figure 2), they include dehydration, malnutrition, asphyxia, aspiration pneumonia, increased length of hospital stays, and, consequently, early mortality. It is worth noting that all variables resulting from oropharyngeal dysphagia are pre-outcome variables, with no confounding factors and no direct causal relationship between oropharyngeal dysphagia and mortality, only with the other studied variables. Thus, saying that dysphagia causes death is not appropriate since it is indirectly related to dysphagia.

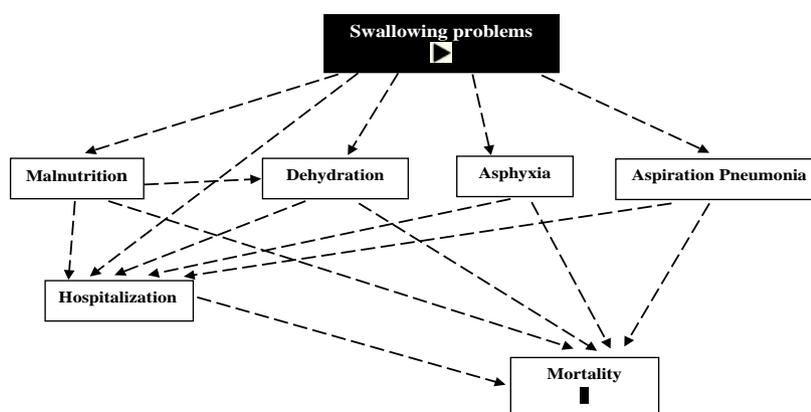


Figure 2: Directed acyclic graph (DAG) to present associations from the literature review.

Legend: Black Background ►: variable exposure; White Background ■: outcome variable; White background: variable prior to the outcome.

IV. DISCUSSION

The process of formulating conceptual systems and converting them into symbolic expressions is called theorizing or constructing theory. The term theory has been defined in various ways by social scientists. A theory consists of one or more functional statements or propositions that deal with the relationship among variables to explain a phenomenon or set of phenomena [70]. The swallowing process and oropharyngeal dysphagia; in other words, any change in the process of eating food between the mouth and the esophagus. Although the theories differ in many ways, we maintain that, in essence, all theories consist of concepts and the relationships between those concepts. A theory is a set of statements about the relationship (s) between two or more concepts or constructions, that is, between the variables we found in the literature and analyzed.

Several criteria have been proposed to evaluate indexed theoretical expressions. If we assume that the purpose of a theory is to help us better understand speech therapy, the primary consideration is whether it offers guidance. According to this perspective, the main criterion for evaluation is utility. Theoretical expressions are valued insofar as they serve as guides for the world we experience. By that, we mean if they allow us to gain some understanding of health practice. If a theory is flawed in some aspect but still provides other unique and insights, it tends to be maintained until something better appears.

The exponential growth in epidemiological studies has reflected an increase in analytical studies [71]. Thus, although theoretical models are used more often than before, the evidence in our research shows that it is still not a robust practice. When dealing with theoretical models in speech therapy research in general, consider whether we should use existing structures as they are, adapt them, or develop new

ones. Speech therapy research and epidemiology often use theoretical frameworks from other areas, such as sociology, psychology, literature, epidemiology and public health, or even basic biological sciences. Health science theories are almost universal and hardly fit perfectly in all subfields, including speech therapy research. We argue that changes in these structures should be supported by theory and not just data.

Much research time has been devoted to the development of scientific models. They are central in many areas to describe the rationale for their modeling approach. This is in line with the concept of models, as they are representations of parts of a whole [72,73]. Different categories of models have been described. For example, an analog model represents of a phenomenon using metaphors and analogies, while a statistical model represents data using mathematical equations. In this article, we are interested in exploratory models, called theoretical models. According to the Stanford Encyclopedia of Philosophy [74], these models are "a starting point of further explanations in which the model is modified and refined" and "provide proofs-of-principle and suggest how-possibly explanations."

Theoretical frameworks are necessary to avoid fishing expeditions (i.e., looking for any associated variable with a significant p-value) and black box epidemiology (i.e., identifying various risk factors that are not always properly connected through a disease theory and considering the mention of the concept of multi-causality sufficient). Theoretical frameworks help to outline data collection and should not be used only for analysis and references. They also help to interpret results in appropriate contexts. Usually, good research starts with a good question. Furthermore, it is generally accepted that good questions can come from experience and observation. This is partially true. Experience and observation can help put a question in

context, but developing a research question is not simple [75], and simple observation can lead to naive questions. Likewise, it is untrue that descriptive epidemiological studies can generate hypotheses [76]. The results of a study without a theoretical framework can easily be misinterpreted due to a lack of important information.

Concerns about the limitations of black-box epidemiology date back to the 1990s [77-79]. Exploratory studies can result in several spurious associations, and these models cannot explain the relationships between variables [80,81]. Traditional epidemiology is tied to proximal risk factors at the individual level and does not consider life course, the complexity of social contexts [82], or methodological challenges [83]. Observational studies are prone to distortions in the selection, confounding, and measurement. Consequently, the validity of the results and conclusions may be at risk. The false-positive of false-negative results can lead to a waste of time. Theoretical frameworks can help improve study design, data collection, analysis, the interpretation of results, and the evaluation of result validity. Without a theory-driven hypothesis, fishing expeditions on large data sets can divert resources from important hypothesis.

Most cross-sectional studies are subject to reverse causality, and it is still uncertain whether dysphagia comes first or health problems such as frailty, sarcopenia, and xerostomia [84,85]. Another factor subject to reverse causality bias is the individual's psychological state, which can be both exposure and outcome. The literature has shown that emotional damage, mainly related to the quality of life and self-perceived health, has been significantly associated with dysphagia [86]. Besides negatively affecting physical health, dysphagia interferes with quality of life. Patients with dysphagia report harmful effects on their social life [87], as they cannot to feed themselves when they eat with family and friends. This results in isolation and depression [88].

Dysphagia is complex and, although it has been increasingly studied, it is important to remember that modern epidemiology has demonstrated that health transcends understanding the health-disease process at the individual level and seeks to view populations [89]. Society is constantly changing, so professionals must update their knowledge about the theoretical and practical strategies for screening, evaluating, and rehabilitating the population in the most beneficial way [23]. Professionals must also consistently improve their knowledge and understanding of the interrelationships between variables and possible triggering outcomes. While not all individual, behavioral, or social health conditions are directly related to dysphagia, they can exert a mediating effect on or confound the causal path [90].

Another important aspect to consider is culture [89]. Cultural diversity requires an understanding of ethnicity, gender, beliefs, and religious issues, along with socioeconomic conditions [91]. The interdisciplinarity between health, culture, and communication implies new paradigms and strategic challenges (political, theoretical, scientific, educational, and clinical). It is essential to fully understand that the disease-health process does not have a single deterministic causal factor, it goes beyond individual health situations and transcends to different levels. Organizational discussions and articulations related to coordinated care are essential to improve individual and collective health [89].

As such, it should be noted that the specific characteristics of each region are reflected in the scope of health services. Regional differences affect public policies and the allocation of health resources [92]. In this context, the literature has two valuable sources of theories about differences in the provision of health services. The first is the inverse care law [93], which states that the availability of health care tends to vary inversely with the needs of the population. The second is the inverse equity hypothesis, which states that any new public health programs and interventions initially reach people of a higher socioeconomic level and increase inequalities between the rich and the poor [94]. Therefore, it is important to consider that having better health care implies greater care, a support network and access to health, the potential for better food and life habits, disease identification and early rehabilitation, as well as access to information [55, 95-97].

It is known that population health is a product of ecological circumstances, resulting from the interaction between human societies with the environment in general, their different ecosystems, and other support processes. It is important to note that populations are heterogeneous and present different social, economic, cultural, technological characteristics at the population level and in the distributions of health and disease. Therefore, incorporate theoretical models so that erroneous conclusions are not added to clinical practice or the sphere of public health. Dysphagia, which continues to emerge as a public health problem, deserves special attention so that health efforts and costs can be properly directed, and adequate diagnoses and therapeutic conduct can be charted from individual measures to public policies in the community.

Authors' contributions: RSR and BNGG conceived the idea, collected data, analyzed data, and wrote and reviewed the final version of the article.

Research center: This study was conducted at the Universidade Federal do Rio Grande do Sul, in Porto Alegre, Brazil.

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