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Cely Carolyne Pontes Morcerf *

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Case Report

Nutcracker Esophagus: Investigation in a Female Patient with Dysphagia and Odinophagy

Cely Carolyne Pontes Morcerf*, Mário Alexandre Matias De Mendonça Filho, José Fernandes Carneiro Rios Junior, Áthila De Almeida Siqueira

Department of Internal Medicine – UNIGRANRIO University – Rio de Janeiro, Brazil

***Corresponding Author:** Cely Carolyne Pontes Morcerf, Department of Internal Medicine – UNIGRANRIO University – Rio de Janeiro, Brazil

Received date: March 02, 2021; **Accepted date:** April 05, 2021; **Published date:** April 09, 2021

Citation: Cely Carolyne Pontes Morcerf, Mário Alexandre Matias De Mendonça Filho, José Fernandes Carneiro Rios Junior, Áthila De Almeida Siqueira., (2021) Nutcracker Esophagus: Investigation In A Female Patient With Dysphagia And Odinophagy. *J. Archives of Medical Case Reports and Case Study.* 4(1); DOI:10.31579/2692-9392/024

Abstract

Introduction: Odinophagia and dysphagia are important symptoms related to the upper gastrointestinal tract, specifically at the level of the oropharynx and esophagus. Objective: to investigate the possible cause of these symptoms through clinical history and complementary exams, thus outlining a more specific approach. Methodology: anamnesis was performed followed by neck ultrasound, fine needle aspiration (FNAP), videolaryngoscopy, computed tomography of the neck, ultrasound with thyroid dopller, esophagogastroduodenal seriography, esophageal manometry, hormone and thyroid antibodies dosage, and evaluation of thyroid antibodies. Speech therapy.

Results: neck ultrasound with topical thyroid gland, diffusible to swallow, diffusely heterogeneous, mixed nodule in upper / middle and anterior third of the left lobe measuring about 2.2cm x 1.6cm x 1.0 cm, in addition to sparse colloidal cysts by the glandular parenchyma, no larger than 0.6 cm, cervical lymph nodes increased in number and dimensions, some coalescent, the largest being located in the left submandibular region, measuring 1.8 cm in its longest axis. US-guided FNAB: Oncotic Cytology- Benign nodule (Category II of the Bethesda system) consistent with benign follicular nodule (Colloid goiter). Paraffin inclusion- Some follicular epithelial ceslls, in addition to leukocytes, amidst eosinophilic background (system I category) Bethesda). High digestive endoscopy without changes. Videolaryngoscopy without alterations. Computed tomography of the neck showing homogeneous prominent palatine tonsils. Heterogeneous thyroid lobes. Submandibular prominent lymph node on the right (IIA) measuring 1.1 cm. Ultrasonography with thyroid doppler confirms the p revious findings. Manometry showed hypertonia of the lower esophageal sphincter, compatible with nutcracker esophagus. Final Considerations: A patient admitted to investigate dysphagia with two months of evolution associated with odynophagia reaches a final diagnosis of nutcracker esophagus, in addition to a benign thyroid nodule. Thus, the investigation should be supplemented on an outpatient basis with high-resolution manometry. He is discharged with a medical prescription and guidance to start monitoring with a gastroenterologist.

Keywords: dysphagia; odynophagia; gastroenterology; nutcracker esophagus

Introduction

Swallowing is divided into the oral phase, which included chewing and forming the bolus, the pharyngeal phase that consists of coordinated processes that perform the closing of the nasopharynx through the veil of the palate, elevation and closure of the larynx, opening of the upper esophageal sphincter and contraction of the pharyngeal muscle. The arrival of the bolus to the esophagus initiates the esophageal phase that transfers the bolus to the stomach by means of contraction and relaxation movements of the esophageal muscles. [1]

Dysphagia is basically defined by the difficulty in swallowing related to the functioning of the structures of the oropharynx and / or esophagus, due to a mechanical, psychogenic or neurogenic pathology. Dysphagia changes the course of food through the upper digestive tract, which can result in an unsafe oral intake resulting in nutritional deficiency, dehydration, aspiration bronchopneumonia and death. It affects all age groups and can be congenital or acquired. In dysphagia, the act of swallowing occurs with difficulty or reduction in the speed of transport of liquid, pasty, solid food or both, and can be located in the oropharynx - defined as high dysphagia - or located in the esophagus - defined as low dysphagia. [1]

Dysphagia is more associated with patients with systemic, neurological or degenerative disorders, which are common during aging. It affects about 20% to 22% of the population over 50 years, reaching rates of 70% to 90% of swallowing disorders in older populations. [2]

The place where patient feels the discomfort is the first place to investigate the cause. The presence or absence of associated symptoms is important to aid diagnosis in some cases. Advances related to dysphagia have occurred, through the understanding of pathophysiology and new methods for examination, favoring a faster diagnosis and the most correct choice for clinical or surgical treatment, with the aim of improving quality of life and reducing complications. [1]

Odynophagia, on the other hand, is defined as pain during the passage of food through the upper digestive tract that particularly affects the pharynx and oscillates from a mild pain, usually retrosternal during swallowing, to pain of extreme intensity. Esophageal injury, neoplastic infiltration, inflammatory processes, ulcers should be investigated and may accompany with secondary dysphagia. [3]

The nutcracker esophagus is described as a manometric abnormality, inserted in the group of primary motor disorders of the esophagus. This abnormality is characterized by the presence of peristaltic waves reaching high amplitudes in the region of the distal esophagus, initially described in studies in cases of patients who presented non-cardiac chest pain. There are still controversies about the meaning of the nutcracker esophagus. The abnormality is considered a variant of diffuse esophageal spasm, consisting of an intense generalized contraction of the esophagus. There is still a strong discussion in the scientific community regarding the real meaning of nutcracker esophagus, however many later studies have recorded the presentation of nutcracker esophagus in patients with dysphagia, in addition to recently being also associated with gastroesophageal reflux disease. [4]

There are few well-defined and reported cases in the literature involving a large number of patients diagnosed with nutcracker esophagus, as well as the number of clinical trials in patients with nutcracker esophagus. We need to pay attention to the importance of suspected diagnosis, the clinical exams and the correct conduct in similar cases.

Fundamentals

2.1 - High Resolution Manometry:

High resolution manometry is the most used technology today for conducting studies of esophageal motility. It allows access to the sphincters and esophageal body giving a simultaneous and panoramic view in a field from the larynx to the stomach. In large research centers, high resolution manometry is gradually replacing conventional manometry with parameters according to the Chicago Classification, which seeks to unify graphical interpretations of high resolution manometry, categorizing esophageal disorders. In relation to conventional manometry, high resolution manometry is faster, more comfortable and has no limitations present in the traditional one, such as moving artifacts.[5]

The relaxation of the lower esophageal sphincter, in conventional manometry, is measured by the minimum pressure, but it can be confused with pseudo relaxation. The high resolution manometry led to the creation of the IRP parameter, which corresponds to the integral of the relaxation pressure, meaning the average pressure of the 4 seconds of greatest relaxation after swallowing in an interval of 10 seconds, which starts at the beginning of swallowing, equivalent to relaxation of the upper esophageal sphincter. [5]

The classification of peristalsis is based on the speed and propagation of the wave, in traditional manometry. In manometry, the distal latency parameter was created, which performs the objective measurement of the peristalsis time, from the beginning of the swallowing process to the epiphrenic ampoule. This parameter occurs due to the interval between the beginning of relaxation of the lower esophageal sphincter and the point of contractile deceleration, which consists of the transition between the esophageal body and the epiphrenic ampoule. The point may be difficult to locate, so a criterion was created in the new version of the Chicago Classification, limiting its location to up to 3 cm from the lower esophageal sphincter, in cases of atypical peristalsis. [5]

Objectives

General Objective

Discuss the importance of diagnostic investigation and correct approaches in patients with dysphagia and odynophagia, for a correct design of specific causes.

Specific Objectives

Perform a bibliographic review on the topics of dysphagia and esophagus in nutcracker, correlating with the clinical findings of the patient.

Describe the clinical history of a patient with complaints of dysphagia and odynophagia, showing the importance of clinical reasoning and esophageal manometry for the final diagnosis of the specific case.

Methodology

Review

At first, it is a descriptive study of the Case Report type, using retrospective information from the patient's medical record, in addition to information obtained directly from the patient's direct medical consultation at the Quinta D'Or Hospital.

Bibliographic Research

Then, a bibliographic search of national and international literature started, covering review articles, original articles and case reports published between 1983 and 2017 and written in English, Portuguese and Spanish. The research was carried out between the months of August and October 2017, on the sites of PubMed (http://www.ncbi.nlm.nih.gov), SciELO - Scientific Electronic Library Online (http://www.scielo.org). The descriptors used were: "dysphagia" and "esophagus in nutcracker", with a final balance of 59 articles. Only articles in Portuguese, English and Spanish were selected, resulting in a number of 30 scientific articles.

Inclusion and Exclusion Criteria

Twenty-one articles were selected based on their relevance, association with the topic, publication in well-known and trusted journals. After reading, articles that did not present adequate methodology or did not address the area of interest were discarded.

Literature Review

Dysphagia Investigation

Swallowing is divided by the oral phase, which included chewing and forming the bolus, the pharyngeal phase that consists of coordinated processes that perform the closing of the nasopharynx through the veil of the palate, elevation and closure of the larynx, opening of the upper esophageal sphincter and contraction of the pharyngeal muscle. The arrival of the bolus to the esophagus initiates the esophageal phase that transfers the bolus to the stomach by means of contraction and relaxation movements of the esophageal muscles. When the difficulty in swallowing is of functional origin and affects the striated muscle it is an oropharyngeal dysphagia and when the smooth muscle is affected it is characterized as esophageal.[6-8].

Dysphagia is classified as oropharyngeal and esophageal and the patient commonly describes the symptom as a sensation of stopping the bolus or sensation of difficulty in passing it somewhere in the neck or sternal region.

A good anamnesis together with the physical examination, are essential to reach a definitive conclusion, since the place where the patient finds the change in the passage of the bolus is the first data to guide during the investigation. Therefore, if the disorder is referred to below the suprasternal region, it should be thought that the dysphagia is of esophageal origin and if it is not, other data should be investigated, as both oropharyngeal dysphagia and esophageal dysphagia can present this location. But the presence of symptoms such as alteration in the formation of the bolus of food, escape of food through the mouth, sialorrhea and fractional swallowing are signs of oral dysphagia. [6]

During physical examination, it is important to evaluate ectoscopy findings that may suggest certain pathologies, such as exophthalmos, tachycardia, tremors and sweat that are related to hyperthyroidism. In addition, changes in the skin, such as erythema on moth wings, telangiectasias, Raynaud's phenomenon, dry skin associated with oropharyngeal dysphagia may suggest scleroderma or other pathologies related to connective tissue. Neurological function should be investigated, if possible by a specialist, to more accurately assess the existence of neuromuscular diseases, Parkinson's disease, myasthenia gravis that can be associated with dysphagia. Respiratory function is important to be investigated to rule out pulmonary complications by aspiration. The exploration of the head and neck may reveal the presence of tumors that can lead to dysphagia and odynophagia and scarring, such as a tracheostomy that may indicate previous treatment of an organic lesion that may be associated with a new picture of dysphagia. [10,11]

However, anamnesis and physical examination do not always close the diagnoses of many patients and complementary examinations end up becoming frequent and necessary to reach the etiological diagnosis. An example is in hypothyroidism where dosages of thyroid hormones are needed. In addition to thyroid hormones, other tests are important because they may be associated with dysphagia, such as: neck ultrasound which can assess the thyroid gland and cervical lymph nodes. Some thyroid nodules depending on the size can compress the esophagus and cause dysphagia. Fine needle aspiration puncture that can assist in the presence of thyroid nodules and histologically define the nodule according to the Bethesda system category. Upper digestive endoscopy that can assess the pharynx, larynx and esophagus and find out if there is an organic lesion that justifies dysphagia. Computed tomography can evaluate the palatine tonsils, lymph nodes. Another exam of great importance is esophageal manometry that measures the strength and function of the esophageal muscles and changes such as sphincter hypertonia for being evidenced in the exam and justifying dysphagia closing the diagnosis. The barium esophagogram can be useful to discover evident organic lesions, such as zenker's diverticulum and plummer-vinson syndrome. Fine needle puncture may be necessary in case of nodular lesions and esophageal manometry to assess the strength and function of the esophageal muscles. [12]

In view of the etiology of dysphagia, treatment consists of resolving the underlying cause. An example of this are patients with esophageal cancer where the treatment will depend on the degree of evolution of the disease that may require from tumor resection, regional lymph nodes with reconstruction of esophagogastric transit to palliative care. It is also necessary to consider the importance of the patient's multidisciplinary approach, with speech therapy being widely used as an aid in the management of many cases. [13,14]

Nutcracker Esophagus

The presence of associated symptoms, such as regurgitation or respiratory symptoms, can consolidate the clinical impression and help in the etiological diagnosis. [9]

The first manometric changes were evidenced and defined in the literature in 1977, in cases of patients who presented angina chest pain and coronary arteriography without changes, with the presence of high amplitude peristaltic waves in the distal esophagus. This abnormality was described and later recognized as nutcracker esophagus, a name preserved until today. [4]

There are still controversies and debate among members of the scientific community regarding the significance of the abnormality. The literature shows that some authors believe that the nut-cracking esophagus is not a cause of disease but a marker of disease. [15]. Several authors also point out the high incidence of the presence of patients diagnosed with nutcracker esophagus and the association with psycho-emotional conditions, with great emphasis on the stress and impairment of the patient's mental health. [16,17]

A retrospective study using a sample space of 97 patients, diagnosed using manometry, with nutcracker esophagus, using as a reference a control group of asymptomatic patients, analyzed the clinical characteristics of the patients, showing a predominance of females with an average age of 50 years, in addition to the evidence of chest pain, dysphagia, heartburn, regurgitation and dyspepsia as the most frequent complaints. Such a study concluded that, despite the existence of controversies associated with the nutcracking esophagus, other complaints are present, in addition to dysphagia and chest pain, even though these are the most commonly encountered. Thus, it is observed the relevance of a study and a thorough analysis of the association of the abnormality with other diseases, such as gastroesophageal reflux, much described in the literature as a very common and important association, together with the investigation of the causes of dysphagia for a correct therapeutic orientation. [18][4]

Case Report

Female patient, 34 years old, with a previous history of Hashimoto's thyroiditis, is admitted to the Quinta D`Or Hospital, on the morning of 10/11/2017, with complaints of dysphagia and odynophagia, reporting having started such symptoms about 2 months, worsening in the last 3 days. She denied other associated symptoms.

Upon examination, in the emergency room, she was lucid and oriented in time and space. Head and neck assessment with palpable, mobile and rubbery right submandibular lymph node, palpable thyroid, mobile to swallow and painful. Remainder of the physical examination without significant changes.

On the day of admission, a computed tomography scan of the neck was requested, where it showed prominent and homogeneous palatine tonsils, heterogeneous thyroid lobes, and a prominent submandibular lymph node on the right, measuring 1.1 cm. Laboratory tests taken that same day had normal results.

Neck ultrasound, fine needle aspiration, videolaryngoscopy, computed tomography of the neck, ultrasound with thyroid dopller, esophagogastroduodenal seriography, esophageal manometry were performed, and thyroid hormone and antibody measurements were also requested, in addition to speech therapy assessment.

Neck ultrasound with topical thyroid gland, mobile to swallow, diffusely heterogeneous, mixed nodule in the upper / middle and anterior third of the left lobe measuring about 2.2 cm x 1.6 cm x 1.0 cm, in addition to sparse colloidal cysts by the glandular parenchyma, no larger than 0.6 cm, cervical lymphododes increased in number and dimensions, some coalescent, the largest being located in the left submandibular region, measuring 1.8 cm in its longest axis. FNA guided by ultrasonography: Oncotic Cytology - Benign

nodule (Category II of the Bethesda system) consistent with benign follicular nodule (Colloid goiter). Paraffin Inclusion - Some follicular epithelial cells, in addition to leukocytes, amid an eosinophilic background (category I of the Bethesda system). Upper digestive endoscopy without changes. Videolaryngoscopy without changes. Computed tomography of the neck showing homogeneous prominent palatine tonsils. Heterogeneous thyroid lobes. Prominent submandibular lymph node on the right (IIA) measuring 1.1 cm. Ultrasonography with thyroid doppler that confirms the previous findings. Manometry showed hypertonia of the lower esophageal sphincter, compatible with nut-breaking esophagus.

As a conduct, clinical support, pasty diet, evaluation by speech therapy, dosage of hormones and thyroid antibodies were performed, in addition to evaluating the performance of esophageal manometry, the main exam to arrive at the patient's definitive diagnosis.

Patient reaches a final diagnosis of esophagus in nutcracker, in addition to benign thyroid nodule. Thus, the investigation will be complemented on an outpatient basis with high resolution manometry. He is discharged with a medical prescription and guidance to start monitoring with a gastroenterologist.

Discussion

Starting the analysis of the case, it is important to highlight the issue of being a female patient, which is the literature shows as the vast majority of the gender that presents nutcracker esophagus. **[15] [18].** The vast majority of studies that report clinical cases compatible with nutcracker esophagus, point to dysphagia as the main target symptom for diagnostic investigation, as in the present case, requiring an active search, through a good anamnesis and request for specific complementary exams for the direction and reasoning of the case, considering differential diagnoses and the presence of other associated pathologies that contribute to the patient's symptoms. Despite all the studies describing cases of nutcracker esophagus pointing to a higher prevalence of females, this fact has no scientific clarification, being just statistical evidence, taking into account the simple frequency. Some studies carried out debate on the existence of a distinction between genders, considering the characteristics of esophageal motility and visceral sensitivity in people without the abnormality. **[16].** However, this fact cannot be considered to justify the prevalence of women with findings compatible with the nutcracker esophagus, since the research carried out only included asymptomatic individuals.

The authors also describe the predominance of a specific age range, around 30 to 50 years, for the symptomatic manifestation in a patient with nutcracker esophagus. When comparing these data with the present clinical case, we observed that the patient is 34 years old, compatible with some cases already reported. The main symptoms that led to the manometry were dysphagia and odynophagia, in the same way as present in the literature, but the patient did not have heartburn or chest pain, which has been described in studies used as a basis for patients with nutcracker esophagus. The literature also shows that a small percentage of patients with nutcracker esophagus present only dysphagia or chest pain as a single symptom, while most patients report two to five symptoms, including dysphagia, odynophagia, heartburn, dyspepsia and chest pain. This fact may suggest that in the first moment of the clinical evaluation, the presence of more than one esophageal symptom would be an alert for the possibility of nutcracker esophagus. However, a broader study is needed, with a considerably large sample space, in addition to comparative analyzes with patients with other motor disorders for this conclusion [4]. The existence of reflux was not verified with the requested complementary exams. The literature points out that many patients with nutcracker esophagus undergo upper gastrointestinal endoscopy and manometric examination for reflux research, thus being able to verify the abnormality of the nutcracker esophagus, and in some cases of associated erosive esophagitis, highlighting the importance of a broad complementary investigation for definition of causes, consequences and abnormal findings in patients complaining of dysphagia and odynophagia. [12] [19] [20] [21]

Studies show that there are generally no abnormalities of the radiological examination in patients with nutcracker esophagus, since the primary peristalsis is preserved in these patients, a fact compatible with the findings of the patient in question. The literature also shows that in patients with a major complaint of dysphagia, motor alterations are more frequent. The study of manometry is essential for the diagnosis and monitoring of these patients. A clinical evaluation carried out with 97 patients with nutcracker esophagus showed that 34% of the patients had abnormal lower esophageal sphincter pressure, being hypotensive in only 12.4% and hypertensive in 21.6%. In that same study, relaxation of the lower esophageal sphincter was incomplete in 16.4% of the patients [4]. In the clinical case in question, the manometry showed hypertonia of the lower esophageal sphincter, corresponding as the main finding present in the literature to manometry in cases of nutcracker esophagus.

Esophageal pH monitoring is also an important test for the diagnostic investigation of the patient in question. One study showed that she had abnormalities in 42% of a group of 52 patients who underwent the test. The first authors who described and evaluated using prolonged esophageal pH monitoring in patients with nutcracker esophagus were ACHEM and collaborators, writing about their findings of an association between Gastroesophageal Reflux Disease and nutcracker esophagus in 35% of patients. The same study showed that 66% of the patients with nutcracker esophagus and with alterations in pH-metry did not have heartburn as the main symptom, but chest pain and dysphagia. [9] [15]. Despite the great advances of today, there is still no definition of how the nutcracker esophagus influences the symptoms of patients.

Final Considerations

National and international literature recognizes the wide variety of clinical manifestations in most patients with nutcracker esophagus, showing the need for a broad diagnostic investigation, from the complete collection of the patient's history to the request for complementary tests that allow identification pathologies and other findings associated with the diagnosis of abnormality of the nutcracker esophagus. Studies also suggest that the nut-cracking esophagus would not be the triggering factor or directly associated with the appearance of these manifestations in the patient, and can be considered as a marker of esophageal disease associated or not with the patient's psycho-affective problems, which lead to psychic changes, worsening the intensity or triggering of the main symptoms. This reinforces the importance of recognizing the nutcracker esophagus to manometry in an initial way and also to outpatient follow-up for this patient, with a preference for the use of high resolution manometry, given the existence of benefits recently described in scientific articles. However, there is still a great controversy about the real meaning of the esophagus in nutcrackers, in addition to the lack of clinical trials and comparative studies with diagnosed patients, thus serving the case of the present work reported and discussed, as a contribution to the characterization of the abnormality, as well as pay attention to the importance of extensive investigation of dysphagia and odynophagia, key points that were essential for the diagnostic suspicion and the manometric finding.

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DOI:10.31579/2692-9392/024