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INDEX

PROSPEROUS AND PRODUCTIVE NEW YEAR

We close the year by presenting to our esteemed readers the latest 2023 edition of the Revista Científica CEREM Goiás. We consider this year a great success, as we managed to contribute significantly to the dissemination of scientific work developed by resident doctors and medical residency services in our region.

With a growing participation of resident doctors and medical residency services, presenting relevant and unprecedented work, we anticipate that the year 2024 will be even more prosperous and productive.

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ACUTE NEGATIVE PRESSURE LUNG EDEMA AFTER EXTUBATION IN RHINOPLASTY: CASE REPORT

FELIPE MENDES FARIA[1]; PEDRO IVO MENESES XIMENES[1]; GUSTAVO SIQUEIRA ELMIRO[1,2]; GIULLIANO GARDENGHI[1,3]

ABSTRACT

Introduction: Negative pressure pulmonary edema (NPPE) is a complication that is rarely described in the literature and has an incidence of 0.1% in patients undergoing general anesthesia, with a higher occurrence in otorhinolaryngological procedures. Pulmonary edema is a complication that appears after anesthesia with glottis closure during the inspiratory period, which significantly reduces intrathoracic pressure. Case report: Individual undergoing elective rhinoplasty. In the operating room with stable vital signs. Subjected to balanced general anesthesia: Pre-oxygenation under a face mask was induced with dexmedetomedine, sulfentanila, ketamine, propofol and atracurium, and periglottic block was performed with ropivacaine and lidocaine. The intraoperative period continued without complications. At the end of the surgery, the patient was extubated in plane due to the onset of intense laryngospasm and coughing. After extubation, the patient developed a drop in oximetry (50%), with the installation of a Guedel cannula and the Jaw-thrust maneuver and with improvement. important aspect of oximetry with supplemental oxygen (O2). Discussion: The formation of non-cardiogenic pulmonary edema has been observed following various forms of upper airway obstruction. The frequency is, however, dependent on the surgical method and procedures. Most of them appear immediately after extubation. The vigorous inspiratory effort against the closed glottis generates negative pressures in the pulmonary interstitium, which favors fluid transudation. The therapeutic approach involves treating the cause with supportive measures, airway permeabilization, administration of supplemental O2 via face mask, bronchodilators and, if necessary, non-invasive, or invasive mechanical ventilation.

KEYWORDS: PULMONARY EDEMA; LARYNGISMUS; RHINOPLASTY; DYSPNEA; OXYGEN

INTRODUCTION

Negative pressure pulmonary edema (NPPE) is a seldom-discussed complication in the literature, with an incidence of 0.1% in patients undergoing general anesthesia. It occurs more frequently in otorhinolaryngological procedures.[1] Pulmonary edema is a complication that arises post-anesthesia with glottis closure during the inspiratory period, leading to increased subatmospheric intrathoracic pressure. This negative pressure promotes transudation of fluid into the alveoli, resulting in the entire clinical presentation of NPPE.[2] Despite few scientific reports in the literature, NPPE is a significant issue in anesthesia practice, with a higher incidence during increased manipulation of the patient's airways, especially in the presence of laryngospasm.1 This article aims to present a case of NPPE, its diagnosis, mechanism of action, and treatment.

CASE REPORT

Female patient, 17 years old, ASA I, with no history of prior surgeries, comorbidities, or drug allergies. She arrives at the surgical center for an elective rhinoplasty at a hospital in Goiânia, Goiás, performed by an otorhinolaryngologist. The patient in question experienced flu-like symptoms one day before the surgery, and the surgeon was informed promptly. Immediate initiation of antibiotic therapy and corticosteroids resulted in a substantial improvement of symptoms on the day of the surgical procedure.

In the operating room, the patient was adequately monitored with pulse oximetry with plethysmographic waveform, electrocardiogram, and non-invasive blood pressure, exhibiting stable vital signs before anesthesia induction. A venous line was established with a 20G needle in the right upper limb. The patient underwent balanced general anesthesia: Preoxygenation with a facial mask using 100% oxygen at 6L/min for 3 minutes. Anesthesia induction was carried out with dexmedetomidine (60 mcg) infused in 250 ml of 0.9% saline, sufentanil (15 mcg), ketamine (20 mg), propofol (120 mg), and atracurium (30 mg). Periglottic block was performed with 10 ml of 0.5% ropivacaine and 1% lidocaine without vasoconstrictor. Tracheal intubation was done with a 7.0 cuffed tube, Cormack-Lehane 2b view, direct and atraumatic laryngoscopy confirming proper tube placement by capnography. Mechanical ventilation was adjusted to maintain an end-tidal CO2 (PETCO2) close to 35 mmHg. Anesthetic maintenance included 1.5-2% sevoflurane with low fresh gas flows of 2L/min on the anesthesia machine and remifentanil target-controlled as per the physician's discretion. Adjuvant medications used were dipyrone 2g, cephalexin 2g, dexamethasone 10mg, ondansetron 8mg, tenoxicam 40mg, and tranexamic acid 750mg.

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The intraoperative period was uneventful and the patient maintained stable vital signs throughout the period. At the end of the surgery, the patient was extubated in a flat plane due to the onset of intense laryngospasm and coughing, and after extubation the patient developed a drop in oximetry (50%). A Guedel cannula was installed and the Jaw-thrust maneuver was performed with a significant improvement in oximetry, 90-92% (with oxygen supplementation). Then, after clinical stabilization, she was taken to the post-anesthesia care unit (PACU), awake, conscious, using a mask with an oxygen reservoir at 7 L/min. After oximetry stabilization, SpO2 of 92-93%, eupneic, she was discharged home. On the same day of hospital discharge, the patient returns to the hospital emergency room due to dyspnea and a drop in O2 saturation (80%) verified by the mother at home. Due to significant clinical worsening and the need for supplemental O2, the patient continued to be admitted to the intensive care unit (ICU). Upon admission to the ICU, already clinically stable, however using supplemental oxygen, the patient underwent laboratory tests, electrocardiogram, viral panel, chest x-ray and chest tomography angiography for diagnostic purposes, the latter being represented by figure 01 bellow:

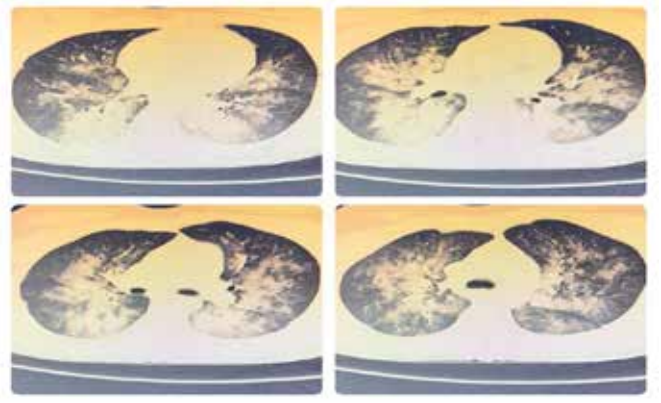

Figure 01: Angiotomography image of the patient's chest (parenchyma) showing confluent acinar and lobular opacities of consolidative and ground-glass nature, at times configuring a mosaic pattern.

The chest angiotomography revealed bilateral and diffuse pulmonary involvement, with a predominance in the right lung. It is characterized by confluent acinar and lobular opacities of consolidative and ground-glass nature, at times forming a mosaic pattern. No filling defects were identified in the evaluated pulmonary arterial branches that could suggest acute pulmonary embolism (PE), as shown in Figure 02. The angiotomography report also mentioned that the pulmonary findings require close clinical and laboratory correlation, considering infectious/inflammatory processes as a hypothesis. To rule out any suspicions of lung disease, the patient underwent a viral panel, COVID-19 test, and other infectious control exams.

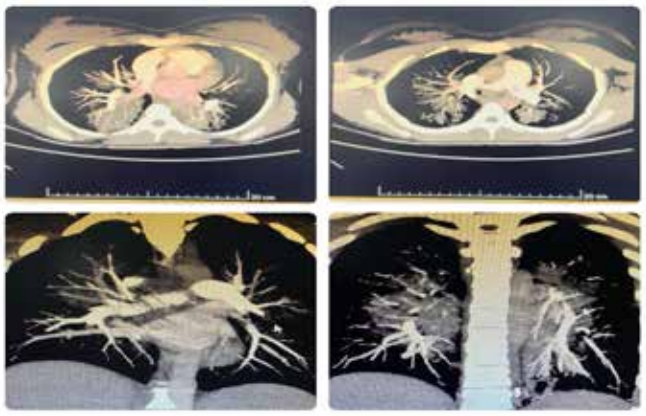

Figure 02: Image of angiotomography of the patient performed upon admission with the objective of ruling out acute pulmonary embolism (PE).

The patient's viral panel was negative for all viruses it included, including Coronavirus, Parainfluenza, Adenovirus, Influenza, Metapneumovirus, Rhinovirus, Bordetella, Mycoplasma, Respiratory Syncytial Virus, and all various subtypes of the mentioned viruses. Among the other conducted tests, it is important to highlight the negative Bacterioscopy and a Procalcitonin level of 0.18 ng/ml. This significantly reduced the possibility of both viral and bacterial infections.

The patient was kept in the intensive care unit (ICU) until clinical stabilization. At the request of the attending physician, a new tomography was performed for added safety before discharge from the ICU, and thus, we obtained the following image:

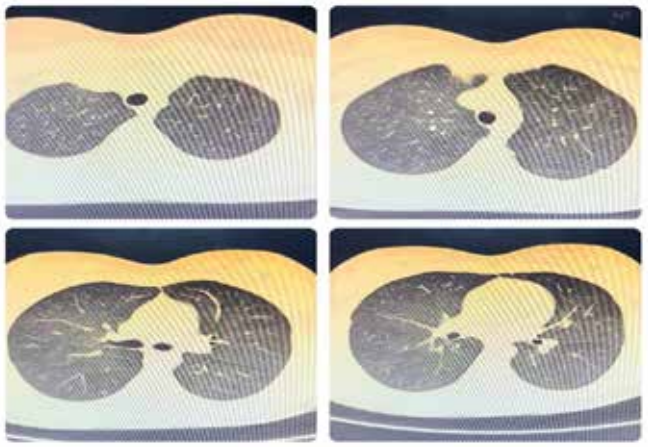

Figure 3: Image of patient, control chest tomography, performed on the 5th day of illness.

The patient's clinical progression in the ICU, demonstrating hemodynamic stability, the absence of respiratory discomfort, and no supplemental oxygen for the past three days with an adequate SpO2 (99%), coupled with a significant improvement in the radiological pattern, led to her

medical discharge from the ICU on 05/09/2023 to a general ward while maintaining favorable parameters. She was ultimately discharged from the hospital on 05/10/2023.

DISCUSSION

The formation of non-cardiogenic pulmonary edema has been observed following various forms of upper airway obstruction, becoming more common in conjunction with laryngospasm (reported in 50% of cases), with an incidence ranging from 0.05% to 5% of anesthetic procedures. The frequency, however, depends on the surgical method, with otorhinolaryngological procedures showing a higher incidence. Approximately 11% of individuals experiencing laryngospasm progress to non-cardiogenic pulmonary edema (NPPE). The latter is a complication that typically occurs with an estimated incidence of 0.1% in general anesthesia practice. [1,3]

The majority of adults who develop acute pulmonary edema (APE) have laryngospasm as the primary triggering factor. The diagnosis is based on clinical manifestations, including ventilatory difficulty followed by inspiratory stridor. Aspiration of a foreign body, bronchospasm, tracheal tube obstruction, laryngospasm, diphtheria, epiglottitis, and strangulation are likely acute causes of non-cardiogenic pulmonary edema (NPPE).[2]

The definition of laryngospasm is the occlusion of the glottis after chemical, mechanical or extrinsically painful stimuli that lead to contraction of laryngeal constrictor muscles. The middle vagus nerve promotes this protective reflex, most of which occurs during an anesthetic emergency, generally in the period prior to extubation. Which makes it possible to increase levels of negative intrapleural pressure, causing APE.[2]

The pulmonary edema, in most cases, manifests immediately after extubation. However, it can occur after a variable period, between two and three hours after airway obstruction, as reported in the second case by Silva et al., where the patient developed dyspnea two hours after extubation. This later manifestation may be explained by an initial protective mechanism. NPPE has a multifactorial physiology. It occurs with forced inspiration against a closed glottis, known as the Müller maneuver, creating a subatmospheric pressure between -50 to -100 mmHg that is propagated to the pulmonary interstitium. This raises venous return to the right side of the heart, resulting in an increase in capillary pulmonary hydrostatic pressure, followed by the transudation of fluid into the alveoli. The negative intrathoracic pressure during left ventricular systole increases afterload, leading to an elevation of end-diastolic volume, reduction of systolic volume, and a decrease in ejection fraction of the left ventricle, promoting an increase in pulmonary vascular pressure. The displacement of the ventricular septum to the left reduces the ejection fraction of the left ventricle (LV) and further elevates microvascular pulmonary pressure due to increased venous return.[2,3,4]

The clinical manifestations include inspiratory stridor, hypoxia, hypercapnia, tachycardia, tachypnea, reduced tidal volume, paradoxical breathing, or uncoordinated ventilatory patterns. Pulmonary edema is accompanied by wheezing and bubbling rales on auscultation, dyspnea, cyanosis, and frothy pink-tinged secretions in the oropharynx.[2]

Complementary diagnostic measures for NPPE include a simple chest radiograph. The typical radiological appearance of this type of edema suggests a predominance of a hydrostatic mechanism. In our case, similar to the case mentioned by Pinhal et al., the images are generally central, bilateral, and located in non-dependent lung zones where the most negative intrathoracic pressures are reached. However, there may be a preferential involvement of a specific lung field. After the regression of the APE, the radiographic pattern may take 12 to 24 hours to return to the previous condition.[2,5]

The screening of symptomatic respiratory diseases is a key element in the preoperative anesthesia assessment. Upper respiratory diseases undergoing general anesthesia carry a higher risk of respiratory complications, including laryngospasm, bronchospasm, and desaturation. However, evidence of lower respiratory tract disease (such as productive cough, wheezing) or systemic disease (such as fever, toxic appearance) constitutes relative contraindications for elective anesthesia. Research suggests that patients with uncomplicated upper respiratory tract infections can undergo general anesthesia without a significant increase in anesthetic complications. Data supporting the risk of perioperative complications related to specific etiological agents are scarce in the literature.[6]

The therapeutic approach involves treating the underlying cause with supportive measures, ensuring airway patency, administering supplemental oxygen through a facial mask, using bronchodilators, and, if necessary, applying non-invasive or invasive mechanical ventilation. Non-invasive ventilation (CPAP/BIPAP) has progressively taken on a prominent role in the treatment of acute respiratory failure in the perioperative period, serving as an effective alternative to invasive ventilation. NPPE, a complication that primarily occurs postoperatively, is recognized in the literature as a well-defined entity but often underdiagnosed, possibly due to its frequently transient and self-limiting course. Invasive monitoring or vasoactive drugs are necessary only in cases of significant hemodynamic changes, which are rare. In the reported case, similar to the majority of cases described in the literature, the patient developed pulmonary edema immediately after extubation, showing significant improvement in dyspnea with the administration of oxygen through a mask, without severe hemodynamic repercussions.[5,7]

CONCLUSION

The present article aims to present a case of NPPE in a young teenager undergoing elective surgery, in which the anesthesiologist reported atraumatic endotracheal intubation with due care for a balanced and higher-quality general anesthesia. The need and importance of continuous support and vigilance with the patient during anesthesia and the time in the post-anesthesia care unit (PACU) were highlighted. It can be concluded that NPPE is still underdiagnosed and underreported in the scientific community. Its treatment involves supplemental oxygen and supportive measures as needed.

Conflicts of interest: The authors have no conflicts of interest to declare.

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CASE REPORT: ANESTHESIA FOR DENTAL PROCEDURE IN A PATIENT WITH SCLERODERMA AND DIFFICULT AIRWAY

ISABELA ALCÂNTARA ROCHA[1], LUCIANA HAHMANN ABREU[1], LARISSA MANZAN DE ALCANTARA BORGES[1], HEBER DE MORAES PENNA[1], GUSTAVO SIQUEIRA ELMIRO[1,2], GIULLIANO GARDENGHI[1,2,3,4]

ABSTRACT

Scleroderma is a chronic autoimmune rheumatic disease that causes progressive sclerosis of the connective tissue and microcirculation, evolving variable levels of tissue fibrosis, which may represent a challenge in procedures that require an advanced airway due to fibrosis of the perioral tissues with limited mouth opening and cervical extension. This article presents the case of a male patient with predictors of difficult intubation due to the physical alterations of systemic scleroderma who underwent multiple tooth extractions under general anesthesia with nasotracheal intubation with fiberscope after intratracheal anesthesia in the awake patient. The intraoperative period was uneventful, and the patient was extubated after effective awakening. The anesthesiologist uses assessments of several clinical variables to predict an airway that is difficult to manage, and the success of the procedure is linked to a thorough evaluation of the patient and the planning of adequate strategies.

KEYWORDS: SYSTEMIC SCLERODERMA; AIRWAY MANAGEMENT; INTUBATION; ORAL SURGICAL PROCEDURES; ENDOTRACHEAL ANESTHESIA

INTRODUCTION

Difficult airway (DA) is defined as a clinical situation in which an experienced anesthetist has difficulty with upper airway ventilation with a face mask, difficulty with endotracheal intubation, or both. The diagnosis is multifactorial, as it depends on the complex interaction between factors intrinsic to the patient, their clinical situation and the level of professional skills.[1]

The DA is responsible for a large part of the complications that interfere with morbidity and mortality linked to anesthesia. The patient's prior assessment, clinical history, semiological data and physical examination provide essential information to predict difficulties and choose the appropriate approach to ensure orotracheal intubation.[2]

One of the main causes for difficult intubation is difficult laryngoscopy, that is, the inability to fully visualize the glottis during direct laryngoscopy, after multiple attempts. The success of the standard intubation technique depends on the ability to manipulate different structures (cervical spine, temporomandibular joint, tissues surrounding the airway). Any condition that alters the constitution or mobility of these structures may represent a level of difficulty in handling.[3]

The risk factors that are proven to be related to the DA and that require early attention are male sex, age over 40-59 years, obesity, diabetes, acromegaly, Obstructive Sleep Apnea Syndrome (OSAS) or rheumatological disease. [4]

Scleroderma is a chronic autoimmune rheumatological disease that causes progressive sclerosis of the connective tissue and microcirculation. It has inflammatory and immunological action, marked by the presence of autoantibodies, and is characterized by variable degrees of tissue fibrosis and small vessel vasculopathy.[5]

Several organs can be affected with an increase in collagen and extracellular matrix proteins, especially the skin, lungs, heart, kidneys and gastrointestinal tract.6 This disease can affect oral and perioral tissues with limited chewing function, skin fibrosis, reduced mouth opening, among other factors that, when combined, can make access to the airway difficult.[5]

Below is a case report of a patient with scleroderma and predictors of difficult intubation, who underwent general anesthesia for dental treatment (multiple tooth extractions). The following report aims to explore possible challenges in the anesthetic procedure, focusing on the possibility of difficult access to the airway in patients with scleroderma.

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CASE REPORT

Male patient, 54 years old, 72 kg, physical status ASA II due to scleroderma, gastroesophageal reflux disease (GERD) and dyslipidemia, controlled with the use of azathioprine, vitamin E, simvastatin, sertraline, formoterol, budesonide, omeprazole and domperidone. In the pre-anesthetic evaluation, the patient presented limited mouth opening of 3 cm (figure 01), reduced cervical mobility (figure 02), Malampatti grade III (figure 03), cervical circumference of 40 cm and positive prayer sign (figure 04), pachydermic skin, normal laboratory tests and low cardiovascular risk. Below are images of the patient's airway assessment.

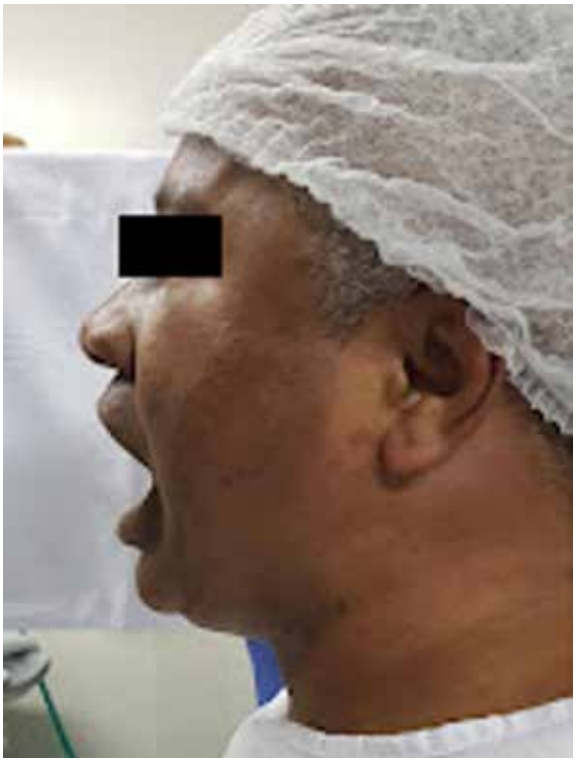

Figure 01 – Maximum mouth opening

Figura 02 - Paciente em extensão cervical máxima em decúbito dorsal horizontal

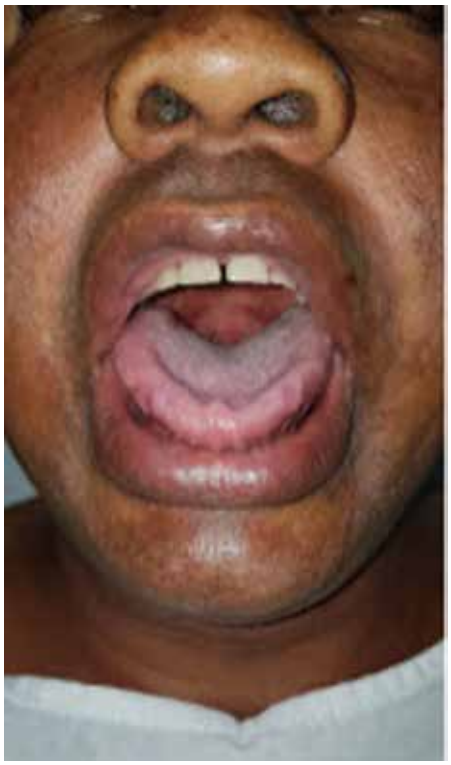

Figure 03 – Malampatti III

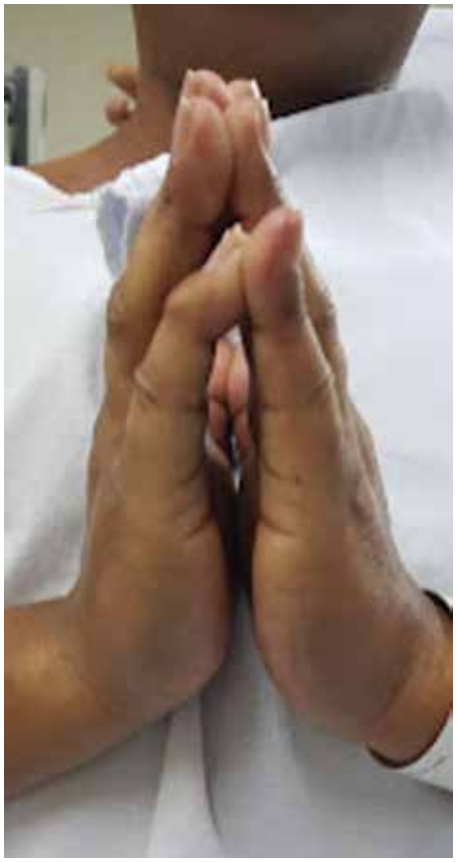

Figure 04 - Positive prayer sign

DISCUSSION

Anesthesiologists use assessments of several clinical variables to predict the difficulty of orotracheal intubation in a patient. However, no isolated clinical measure or test performed at the bedside is capable of fully excluding the possibility of a difficult airway and the assessment of a set of criteria presents greater sensitivity than the evaluation of a single isolated criterion. According to the systematic review by Detsky et al, a previous history of difficult intubation was the biggest risk factor for predicting future difficult intubation and the best bedside test that can be performed alone to identify a difficult airway is the upperlip bite test (class greater than or equal to 3 with specificity of 0.96 for difficult intubations). Other tests such as reduced mobility, Malampatti greater than or equal to 3, mouth opening less than 2.5 cm and positive prayer sign were moderately accurate in predicting a difficult intubation. [7]

Scleroderma may be related to worse scores in clinical assessments of difficult airway predictors because it is an autoimmune disease that affects several organs, including the skin and subcutaneous tissue. There are two courses of the disease: diffuse cutaneous scleroderma, which is characterized by the rapid development of symmetrical skin thickening of the proximal and distal parts of the limbs, face and trunk; and limited cutaneous scleroderma, defined by symmetric thickening of the skin limited to the distal parts of the limbs and face. According to the literature, small airway dysfunction is more common in patients with limited cutaneous sclerosis compared to the more severe diffuse cutaneous sclerosis.[8,9]

Airway management can be a problem in patients with scleroderma. Facial involvement includes a smooth, wrinkle-free forehead, tight skin over the nose, shrinkage of the tissue around the mouth and perioral radial grooves, thinning of the lips and microstomia, the nose takes on a pinched appearance, the affected skin becomes hardened, smooth and adhered to the underlying structures, often with hyper- and hypopigmentation, hairless, dry and thick. The limitation of mouth opening is probably related to fibrosis of the temporomandibular joint, consequently leading to a worse Malampatt score. Fibrotic changes in the neck can cause limited cervical extension, which is an indicator of difficulty in mask ventilation and the need for fiberoptic bronchoscopy-assisted intubation. Therefore, patients with scleroderma, especially in advanced stages, have a high probability of having a difficult airway. [8.9]

In the literature, few studies address specific guidelines regarding the perioperative care of patients with scleroderma, but attention must be paid to its peculiarities, which extend beyond the predictors of a difficult airway. Care must be taken when approaching the airway of patients with scleroderma, as the risk of aspiration is higher than that of the general population. This is due to the fact that 90% of patients with scleroderma have GERD and other gastrointestinal dysmotility disorders. Gastric involvement and hypotonic tone of the lower esophageal sphincter result in impaired emptying and food stasis. There is a high prevalence of esophageal dilation and may be related to an increased risk of centrilobular fibrosis of the lung. Therefore, pre-operative preventive measures must be adopted, such as the administration of antihistamines (H2) and, when appropriate, pre-operative nasogastric aspiration. This recommendation is particularly strong, as aspiration events have an independent association with risk of death in hospitalized patients with sclerosis. The use of a nasogastric tube must be indicated with caution, as it further increases the risk of esophageal perforation, due to the high incidence of esophageal stenosis. [9.10]

Recognizing the risk of difficult mask ventilation and endotracheal intubation, rapid sequence intubation may not be advisable, and the risk of aspiration should be considered before induction of anesthesia. Furthermore, correct positioning and padding of bone ends must be carried out due to the potential risk for peripheral neuropathies.9

There are reports of incidental findings of difficult intravenous access due to skin thickening. Intravenous access can pose another challenge to the anesthetist, and a low threshold is prudent to guide the insertion of the vascular catheter using ultrasound.[9]

Reconhecidas as particularidades do paciente, deve ser realizado o planejamento do ato anestésico, desde a monitorização, punção do acesso venoso, cuidados com a via aérea, o intra e pós operatórios. Deve-se ter uma estratégia pré-formulada para o manejo da via aérea difícil prevista, para garantir um desfecho favorável e a segurança do ato anestésico. No caso clínico em questão, foi optado por intubação com o paciente acordado devido a suspeita de intubação difícil, ventilação difícil (máscara facial/via aérea supraglótica), e aumento do risco de aspiração, seguindo as diretrizes de prática da Sociedade Americana de Anestesiologistas para o manejo da via aérea difícil.[11]

Once the patient's particularities are recognized, planning of the anesthetic procedure must be carried out, from monitoring, venous access puncture, airway care, intra and post-operative care. There must be a pre-formulated strategy for managing the predicted difficult airway, to ensure a favorable outcome and the safety of the anesthetic procedure. In the clinical case in question, intubation was chosen with the patient awake due to suspicion of difficult intubation, difficult ventilation (face mask/supraglottic airway), and increased risk of aspiration, following the practice guidelines of the American Society of Anesthesiologists for difficult airway management.[11]

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CASE REPORT OF NEURO-BEHÇET'S DISEASE

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ABSTRACT

The neurological disease of Behçet's is a manifestation of Behçet's Disease, a rare condition related to autoimmune vasculitis. It causes recurrent inflammation in various parts of the body, including mouth, genital organs, skin, eyes, and in some cases, neurological manifestations. Neurological symptoms vary and can include encephalitis, meningitis, cerebral venous thrombosis, hemorrhages and cerebral aneurysms. Here's a case report of a 59-year-old male patient presenting clinic and radiologic presentation of Limbic Encephalitis with excellent response to imussupression treatment.

KEYWORDS: BEHÇET'S DISEASE; LIMBIC ENCEPHALITIS

INTRODUCTION

Behçet's Disease (BD) is an inflammatory condition characterized by involvement of large, medium, and small blood vessels. Disseminated vasculitis is believed to be the main trigger for systemic manifestations, including recurrent oral ulcers associated with ocular, urogenital, neurological, cutaneous, gastrointestinal, and articular symptoms [1].

When a patient with confirmed Behçet's Disease develops neurological symptoms, it is diagnosed as Neuro-Behçet's Disease. Involvement of the central nervous system is observed in about 9% of individuals with BD, more commonly in males [2].

It can affect both the brain parenchyma and the vascular structures of the central nervous system (CNS) in its non-parenchymal form. Known consequences of Neuro-Behçet's disease include recurrent meningoencephalitis, cranial nerve paralysis, epilepsy, cerebral venous thrombosis, and episodes of diencephalic and brainstem dysfunction that may mimic strokes [2].

The presentation can be acute, with meningoencephalitis or cerebral venous thrombosis, or progressive, with dementia, ataxia, and dysarthria. Cognitive impairment can occur independently of neurological manifestations [3].

CASE REPORT

A male patient, 59 years old, was referred to the Neurology Institute of Goiânia due to abdominal pain associated with headaches and lesions on the tongue. Treatment with antibiotics (Amoxicillin and Clavulanic Acid) was initiated. The patient presented acute memory impairment, with frequent repetitions due to difficulty in retaining new information, starting two days before. In a symptomatological interview, a history of long-standing recurrent headaches was evident, as well as frequent facial pustules and oral aphthae (tongue and buccal mucosa) – in the last two years, he had monthly ulcerative lesions. The patient reported a subacute episode of imbalance, diplopia, and paraparesis five years before, diagnosed at the time with ischemic pontine stroke. He underwent extensive investigation, including tests for vasculitis, but no definitive etiology was found. Retrospective evaluation of cranial images revealed bilateral lesions in the pons that did not respect vascular territories, with a tumefactive effect, vasogenic edema, and contrast enhancement within the lesion suggestive of an inflammatory pattern. Additionally, he had a diagnosis of dyslipidemia and was taking pantoprazole, simvastatin, and aspirin. On physical examination, there was a slight alteration in memory, especially episodic memory, without other focal neurological deficits. The patient reported a fall in the kitchen four days before. Ectoscopy of the left elbow showed prominent inflammatory signs, with significant swelling and redness. During hospitalization, imaging and laboratory tests were requested to clarify the presented pathology. On the first day of hospitalization, Magnetic Resonance Imaging (MRI) of the skull and a neoplastic/infectious screening with Computed Tomography (CT) of the Chest and Abdomen were performed. Cranial MRI showed a lesion with hypersignal and significant edema in the left hippocampal formation, sparing the amygdala, with heterogeneous and irregular leptomeningeal enhancement from the ependymal plexus of the left lateral ventricle and diffusion restriction. There was

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also a band-like lesion in the central region of the pons, with hyposignal in T1, hypersignal in T2, without contrast enhancement or diffusion restriction (Figure 1). This involvement characterizes a clinical and radiological picture compatible with Limbic Encephalitis.

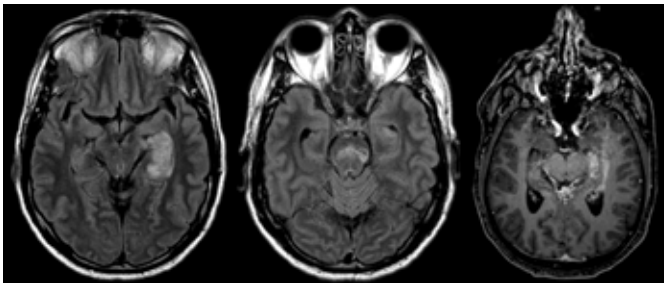

Figura 1: Cranial magnetic resonance imaging. (A) Axial FLAIR, showing vasogenic edema and tumefactive effect involving the left hippocampal formation; (B) Axial FLAIR, demonstrating signal alteration in the pons, bilaterally transversely (band-like), with cavitations, suggestive of a sequela; (C) Axial post-contrast T1, highlighting leptomeningeal and cortical enhancement of the hippocampal formation and contiguous enhancement of the left choroid plexus.

Due to significant inflammation in the left upper limb, the patient underwent an elbow MRI, which revealed olecranon bursitis associated with signs of infectious/inflammatory process around it and a focus of bone edema in the olecranon. Drainage of the collection was performed, and antibiotic therapy was escalated to Ceftriaxone. Abdominal CT showed supernumerary mesenteric lymph nodes. Chest CT showed no alterations. Lumbar puncture (LP) was performed, and the results showed significantly increased cellularity – 348 cells (55 lymphocytes, 15 plasma cells, and 23 monocytes); elevated protein levels (160); and increased lactate of 3.6 mg/dL.

On the second day of hospitalization, additional investigative measures included a normal Electroencephalogram (EEG). Additional laboratory tests performed included: anti-nuclear antibody (ANA) with a fine dense speckled pattern 1:160, non-reactive Anti-DNA, Anti-Sm < 0.7 U/mL, C4 38 mg/dL, Anti-Ro < 0.3 U/mL, C3 228 mg/dL, rheumatoid factor < 20 IU/mL, non-reactive ANCA, and Angiotensin-Converting Enzyme (ACE) 35 U/L, all within normal ranges, except for a significantly elevated erythrocyte sedimentation rate (ESR) with a value of 82.

On the third day, the Pathergy test was performed – the insertion of a 5 mm needle into the forearm, with readings at 24 and 48 hours. The result was strongly positive, with the appearance of a 6 mm papule. With this, the diagnosis of Neuro-Behçet's disease was established, considering the following diagnostic criteria (recurrent oral aphthous ulcers, pseudofolliculitis, and papulo-pustular lesions, positive Pathergy test). Immunosuppressive therapy was then initiated, consisting of methylprednisolone pulse therapy for three days, followed by a course of Cyclophosphamide.

During the 10-day hospitalization, the patient's ESR (erythrocyte sedimentation rate) decreased from 82 on the first day to 23 on the 9th day, and the PCR (C-reactive protein) was 8.7 at discharge. The patient was discharged without headaches, with complete improvement of oral aphthae, resolution of inflammation in the left elbow, and the amnestic picture.

DISCUSSION

Behçet's neurological disease is defined as a set of neurological signs and symptoms in patients with confirmed Behçet's disease (BD). It includes a variety of presentations, primarily involving the central nervous system and, more rarely, the peripheral system. The latter is characterized by neuropathies and myopathies, often in a subclinical manner and confirmed by electrophysiological studies [2].

Manifestations related to the central nervous system (CNS) can be divided into parenchymal and non-parenchymal, with the former being more common and likely affecting the patient in question. Truncal, hemispheric, spinal syndromes, and meningoencephalitis can be clinical manifestations of this subtype. Non-parenchymal involvement includes arterial disorders and venous sinus thrombosis. [4].

Patients with Behçet's disease have a higher risk of experiencing cardiovascular events, such as heart attack and stroke, with the latter being considered a differential diagnosis, especially in patients over 50 years of age [5]. Symptoms such as headache and polyneuropathy can occur secondary to Behçet's disease itself and the medications used to control the disease. In most cases, they represent an underlying uncontrolled inflammatory condition [2].

The diagnosis of Behçet's Disease is based on diagnostic criteria according to the International Study Group (ISG) diagnostic criteria published in 1990 [6], with a new proposal in 2014 that included an expansion of diagnostic criteria to improve sensitivity and specificity [7]. The Pathergy test, a non-mandatory criterion but scoring in favor of the diagnosis, involves skin hyperreactivity followed by trauma with a needle. This test has high specificity, although there are reservations regarding its inconsistent reproducibility and variable sensitivity. The Pathergy phenomenon may be evidence of the endothelial dysfunction characteristic of vasculitis, a group that includes BD [8].

It is important to differentially diagnose the condition from Multiple Sclerosis, Systemic Lupus Erythematosus, Sarcoidosis, CNS infections, and other causes of inflammatory pathologies. Encephalitis, encephalomyelitis, and meningoencephalitis share symptoms with BD, such as headaches, reduced level of consciousness, behavioral changes, and epileptic seizures. Nonetheless, it poses a challenge for neurologists as it relies on the summation of diagnostic criteria [4].

The case report in question not only presents a rare dis-

ease but also brings an extremely unusual clinical and radiological presentation for BD. Limbic encephalitis does not represent a described presentation in the disease, especially with exclusive hippocampal involvement associated with leptomeningeal enhancement with venous pattern restriction, making this report essential for medical literature.

The treatment of the neurological manifestation of BD aims to control the patient's inflammatory condition. The main approach involves the use of intravenous methylprednisolone, followed by oral corticosteroid therapy. Cyclophosphamide, Azathioprine, Methotrexate, and immunobiologics such as Infliximab may be used as indicated, usually required as maintenance therapy [7].

CONCLUSION

The case of the presented patient illustrates the complexity and diversity of DB manifestations, especially when there is an atypical presentation such as limbic encephalitis. Early identification of Neuro-Behçet's Disease and differentiation between its parenchymal and non-parenchymal manifestations are crucial for proper treatment. In this specific case, the patient's favorable response to pulse therapy and subsequent treatment is encouraging and highlights the importance of early diagnosis and appropriate management of the condition. However, long-term follow-up is essential since DB is chronic and requires continuous monitoring to prevent relapses and complications. Ultimately, the case underscores the importance of awareness and education about DB, especially its neurological manifestations, to ensure early diagnosis and effective treatment, thereby improving the quality of life for patients affected by this rare and challenging condition.

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BRADYCARDIA-TACHYCARDIA SYNDROME IN A POST-OPERATIVE CARDIAC SURGERY PATIENT

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ABSTRACT

The bradycardia-tachycardia syndrome is a condition characterized by episodes of supraventricular tachycardia followed by sinus bradycardia, resulting in significant heart rate variability. This syndrome can be associated with various cardiac abnormalities, including valve abnormalities and atrial dysfunction. The presented case is of a patient who sought emergency medical attention due to episodes of dyspnea with minimal exertion, orthopnea, and paroxysmal nocturnal dyspnea, associated with lower limb edema. The patient had a mechanical mitral prosthesis due to decompensated heart failure and prosthetic leaflet fracture, with three reoperations. During the physical examination, irregularities in the heart rhythm were observed, and a transthoracic echocardiogram (TTE) revealed moderate tricuspid regurgitation, moderate biatrial enlargement, and estimated pulmonary arterial hypertension of 80 mmHg. Continuous cardiac monitoring recorded prolonged pauses in the context of episodes of supraventricular tachycardia followed by sinus bradycardia, characteristic of bradycardia-tachycardia syndrome. Additionally, a normal ventricular ejection fraction, moderate biatrial enlargement, and pulmonary arterial hypertension were observed. Bradycardia-tachycardia syndrome can lead to hemodynamic changes and is associated with adverse cardiovascular risks. Treatment includes pharmacological control and, in some cases, lung transplantation. In this case, the patient received outpatient pharmacological treatment and underwent pacemaker implantation.

KEYWORDS: RADYCARDIA-TACHYCARDIA SYNDROME; TRANSTHORACIC ECHOCARDIOGRAM; TRICUSPID REGURGITATION; PULMONARY ARTERIAL HYPERTENSION

CASE REPORT

A female patient, 51 years old, with a history of heart disease, sought emergency medical services after mentioning that, over the past 14 days, she had been experiencing periods of dyspnea with minimal exertion, orthopnea, and paroxysmal nocturnal dyspnea, associated with lower limb edema. The patient had a mechanical mitral prosthesis due to decompensated heart failure, as well as a prosthetic leaflet fracture, for which she had undergone three reoperations.

During the physical examination, an irregularity in the cardiac rhythm was observed, and a transthoracic echocardiogram (TTE) was requested. The TTE results showed normal functionality of her mechanical mitral prosthesis and preserved biventricular systolic function. However, moderate tricuspid regurgitation, moderate biatrial enlargement, and an estimated pulmonary arterial hypertension of 80 mmHg were noted.

During the continuous 24-hour cardiac monitoring, a total of nine prolonged pauses were recorded, with an average duration of 6.2 seconds (Figure 1). These pauses were observed in the context of episodes of supraventricular tachycardia followed by sinus bradycardia, characteristic of bradycardia-tachycardia syndrome (Figures 2 and 3).

The patient reported that during the pauses, she experienced a sensation of imminent fainting, accompanied by dizziness and irregular palpitations. The episodes occurred unpredictably, significantly impacting her quality of life.

Figure 1. Bradycardia-tachycardia syndrome. Nine pauses are observed, with higher frequency between 5 and 6 am.

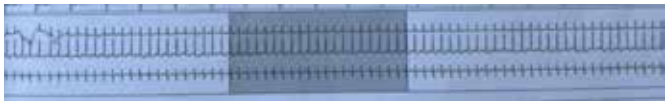

Figure 2. Bradycardia-tachycardia syndrome. A supraventricular tachycardia (SVT) is observed. The maximum recorded frequency in this segment is 156 bpm.

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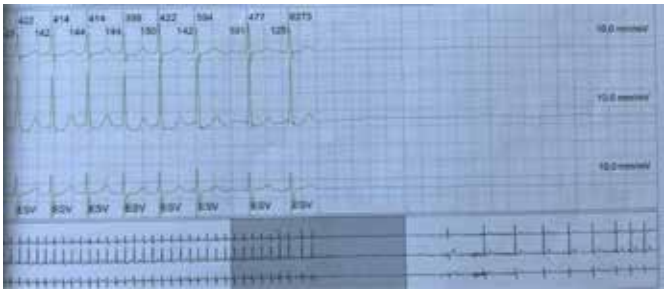

Figure 3. Bradycardia-tachycardia syndrome. In the three observed lines, there is a non-sustained supraventricular tachycardia (SVT) that stops spontaneously. In the first line of the record, a longer sinus pause of 6.2 seconds is observed, followed by sinus bradycardia, with a gradual recovery of the normal sinus node heart rate. The bradycardia is a result of the preceding tachycardia, which decreased the automatic activity of the sinus node.

In the TTE, the prolonged tracing of lead D2 revealed a supraventricular tachycardia that abruptly ceased, followed by a sinus pause and subsequent sinus bradycardia, with gradual recovery of the heart rate. Considering that bradyarrhythmia seemed to be the cause of the syndrome, the medical team diagnosed the patient with sinus node pathology characteristic of bradycardia-tachycardia syndrome and decided to admit her for further investigation of the arrhythmia and the implantation of a cardiac pacemaker.

Continuing with the results, the patient had mild prosthetic regurgitation. The patient's ventricular ejection fraction was 65%, with moderate biatrial enlargement. The normal systolic and diastolic diameters of the left ventricle were 30 mm and 47 mm, respectively, while in the right ventricle, the diastolic diameter was 27 mm. The patient had normal dimensions of the aortic root (31 mm), while the left atrium had a diameter of 47 mm.

After undergoing cardiac pacemaker implantation, the evaluated patient showed improvement in her overall condition. She was breathing normally without discomfort and was undergoing respiratory and motor physiotherapy.

DISCUSSION

The bradycardia-tachycardia syndrome is a condition characterized by episodes of supraventricular tachycardia followed by sinus bradycardia, resulting in significant variability in heart rate. This syndrome can be associated with various cardiac abnormalities, including valvular abnormalities and atrial dysfunction.[1] In this case report, the patient with bradycardia-tachycardia syndrome presented with moderate tricuspid insufficiency, moderate biatrial enlargement, and pulmonary arterial hypertension.

The moderate tricuspid regurgitation observed in this case indicates impairment of the tricuspid valve function, which separates the right atrium from the right ventricle. This type of regurgitation may be related to structural or functional changes in the valve, such as dilation of the valve annulus, rheumatic disease, endocarditis, or right ventricular dysfunction. Scientific literature has shown that moderate tricuspid regurgitation is associated with an increased risk of adverse cardiovascular events, such as heart failure, arrhythmias, and cardiovascular mortality.[2]

The moderate biatrial enlargement observed in this case suggests dilation of the right and left atria. Atrial dilation may be related to volume or pressure overload, as seen in cases of heart failure, valve diseases, pulmonary arterial hypertension, or arrhythmias. In the patient's case, her medical condition as a carrier of bradycardia-tachycardia syndrome may explain this event. Literature indicates that biatrial enlargement is associated with a higher risk of atrial fibrillation, thromboembolic events, and cardiovascular complications.[3]

A previous study by Choi et al.4 (2015) revealed that patients with bradycardia-tachycardia syndrome tend to have an ejection fraction greater than 63%, with manifestations of eccentric left ventricular hypertrophy and increased left atrium observed through transthoracic echocardiography. Cardiomegaly can also occur and is observed in chest radiographs of patients with bradycardia-tachycardia syndrome.

The patient in this case report had estimated pulmonary arterial hypertension of 80 mmHg, with an estimated pulmonary artery systolic pressure of 56 mmHg. Pulmonary arterial hypertension can result from various causes, such as heart disease, chronic lung disease, pulmonary embolism, or genetic diseases. Elevated pulmonary arterial pressure is associated with increased workload on the right side of the heart, leading to complications such as right heart failure and reduced survival.[5,6]

The bradycardia-tachycardia syndrome can lead to hemodynamic changes in the heart and blood vessels, contributing to an increase in pulmonary artery pressure. During tachycardia episodes, the heart may have difficulty pumping blood adequately to the lungs, resulting in an increase in pressure in the pulmonary arteries. Additionally, subsequent bradycardia can lead to a decrease in cardiac output, contributing to an increase in pulmonary arterial pressure.[6]

The treatment of pulmonary arterial hypertension generally involves specific pharmacological approaches, and in some cases, lung transplantation may be considered.[7] The patient in question underwent outpatient treatment with Losartan (50mg), Aldactone (25mg), Aspirin (100mg), furosemide (40mg), and metoprolol (50mg). Additionally, for antibiotic prophylaxis, cefuroxime (750mg) for three days, Tazocin, and Meropenem were prescribed.

According to studies, pharmacological control in patients with bradycardia-tachycardia syndrome can be used to regulate ventricular frequency. Medications such as amiodarone, diltiazem, verapamil, and digoxin may be prescribed, and atrioventricular nodal conduction may be delayed to adjust the ventricular response.[8,9] However,

since such treatments can worsen bradyarrhythmia, it is necessary to insert a pacemaker before attempting pharmacological control.[10] In cases where bradycardia is not a result of drug therapy (such as digitalis or beta-blockers), abnormal sinus node physiology should be considered.[4]

It is worth noting that the presence of bradycardia in atrial disease itself may predispose to the emergence of supraventricular tachycardia. The hemodynamic effects of bradycardia are more significant than those of tachycardia. It is important to emphasize that the supraventricular tachycardia that promotes bradycardia should not involve the use of antiarrhythmic medications, as these may more intensely suppress the dysfunctional sinus node. In the present case, the patient underwent the implantation of a cardiac pacemaker, as recommended by the literature.[7,10,11,12]

CONCLUSION

The report of this case allows us to conclude that bradycardia-tachycardia syndrome is a characteristic arrhythmia of sinus node disease. This case report highlights the importance of identifying and appropriately treating bradycardia-tachycardia syndrome, with therapeutic approaches such as pacemaker implantation and pharmacological therapy. The effective management of this condition can improve the patient's quality of life, reduce the risk of cardiovascular complications, and promote favorable clinical outcomes.

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CASE REPORT OF PACEMAKER IMPLANTATION IN A PATIENT WITH ANOMALOUS DRAINAGE OF THE SUPERIOR VENA CAVA

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ABSTRACT

Background and objective: Persistent left superior vena cava (PLSVC) is a rare embryologic remnant anomaly. Mostly both superior vena cava coexist, however if there is Agenesis of the Right Superior Vena Cava (ARSVC) the venous drainage to the heart will be made to the right atrium, through the coronary sinus. Usually asymptomatic, this malformation can be detected when patients undergo procedures involving the superior vena cava. This is a case report of a patient with PLSVC and ARSVC who underwent pacemaker implantation with greater technical difficulties due to lack of preparation for the anatomical anomaly, as well as the difficulty in handling sedoanalgesia Case report: Male patient, 65 years old, with Coronary Disease, 5 years before undergoing myocardial revascularization, with 1st degree atrioventricular block (AVB). Admitted for permanent cardiac pacemaker implantation. He was admitted to the Surgical Center and received monitoring, venipuncture with a 20 G catheter, and sedoanalgesia with fentanyl and propofol was initiated. Venous puncture was performed in the right subclavian vein, but when checking the position of the guide wire, an anomalous ventricular pathway was visualized. After multiple repositioning attempts, the anomalous intraventricular pathway persisted. During manipulation, the patient presented supraventricular extrasystoles and increased demand for sedatives. When revisiting preoperative exams and found Angiotomography of the Coronary Arteries with a report of "probable PLSVC and ARSVC with anomalous drainage into the coronary venous sinus".

KEYWORDS: PERSISTENT LEFT SUPERIOR VENA CAVA; ARTIFICIAL PACEMAKER; CONSCIOUS SEDATION

INTRODUCTION

The persistence of the left superior vena cava (PLSVC) is a rare and sparsely described embryological remnant anomaly in the literature. Its incidence varies from 0.1 to 0.3% [1] in healthy adults to 4.3% in patients with congenital heart diseases [2]. Mostly, both superior vena cavas coexist, but if there is embryological regression and degeneration of the right anterior cardinal vein, it will result in the absence of the right superior vena cava, and venous drainage to the heart may be carried out by the left superior vena cava into the right atrium through the coronary sinus [2]. Due to the frequency of asymptomatic carriers, this malformation is often incidentally detected when patients undergo central venous catheter placement, pacemaker implantation, or open-heart surgery [3].

The embryological venous system has, as its essential structures, the cardinal veins. The upper and lower cardinal veins combine to form the duct of Cuvier, which drains into the bicarotid venous sinus and develops into the right atrium. The duct of Cuvier, along with the caudal part of the right superior cardinal vein, will form the right superior vena cava (RSVC), while the left common cardinal vein and the caudal part of the left superior cardinal vein will regress [3]. If this regression does not occur, it results in PLSVC [3]. One hypothesis to justify this event involves various embryological conditions leading to a reduction in the size of the left atrium. Consequently, it will not have the necessary dimensions to compress the coronary sinus and the left cardinal vein, culminating in PLSVC [3].

This is the report of a case of a patient with PLSVC undergoing pacemaker implantation due to atrioventricular block (AVB), with the discovery of agenesis of the right superior vena cava (ARSVC), resulting in greater technical and analgesic management challenges.

CASE REPORT

Male patient, 65 years old, with a history of coronary artery disease, underwent myocardial revascularization 5 years ago, and currently has first-degree atrioventricular block (AVB) while on aspirin 100 mg/day and enalapril 20 mg/day. The patient was admitted for the implantation of a permanent cardiac pacemaker. In the pre-anesthetic evaluation, a complete blood count, renal function tests, 12-lead electrocardiogram showing first-degree AVB, and

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transthoracic echocardiography revealing concentric ventricular hypertrophy with preserved ventricular function were assessed. The chosen anesthetic technique was light sedation and local anesthesia. Upon entering the operating room, the patient was monitored with pulse oximetry, non-invasive blood pressure, and cardiac monitoring. Cardiac monitoring showed the presence of first-degree AVB with a heart rate of 42 beats per minute, maintaining adequate capillary filling time and blood pressure. A 20 G catheter was inserted for venous access, and sedation was initiated with a bolus of 50 mcg fentanyl and continuous infusion of Propofol using the Schnider model, with a target concentration of 1.0 microgram/ml. After sedation, local anesthesia with 100 milligrams of lidocaine was administered, followed by a right infraclavicular incision and venous puncture under direct visualization of the right subclavian vein. During the advancement of the guide wire, there was no resistance. However, when checking the position of the guide wire through radiographic images, an anomalous ventricular trajectory was visualized (Figure 01).

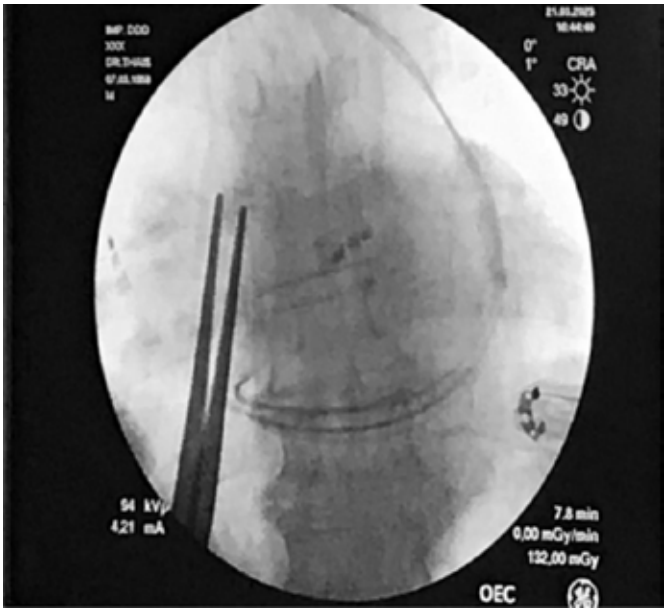

Figure 01: radiological image taken using a surgical arch showing the positioning of the electrodes.

After multiple attempts to reintroduce it into the proper position, the decision was made to change the puncture site. Ultrasound-guided venous puncture was performed in the left subclavian vein. During the introduction of the guide wire, resistance was encountered. Upon checking the X-ray, it was observed that even with the change in the puncture site, the anomalous intraventricular trajectory persisted. During the frequent manipulations, the patient experienced multiple episodes of supraventricular extrasystoles due to the introduction and reintroduction of intracavitary wires, in addition to an increased demand for sedatives to maintain adequate sedoanalgesia, considering the prolonged painful stimulus and time.

The responsible cardiologist decided to revisit preoperative exams and found coronary artery angiotomography (figure 02) with a report of "probable ARSVC agenesis and PLSVC with anomalous drainage into the coronary sinus." Keeping in mind the diagnosis of PLSVC, they resumed the procedure at the initial site and repositioned the electrodes, requiring multiple stimulus tests to find the optimal positioning for both electrodes and, consequently, for the pacemaker.

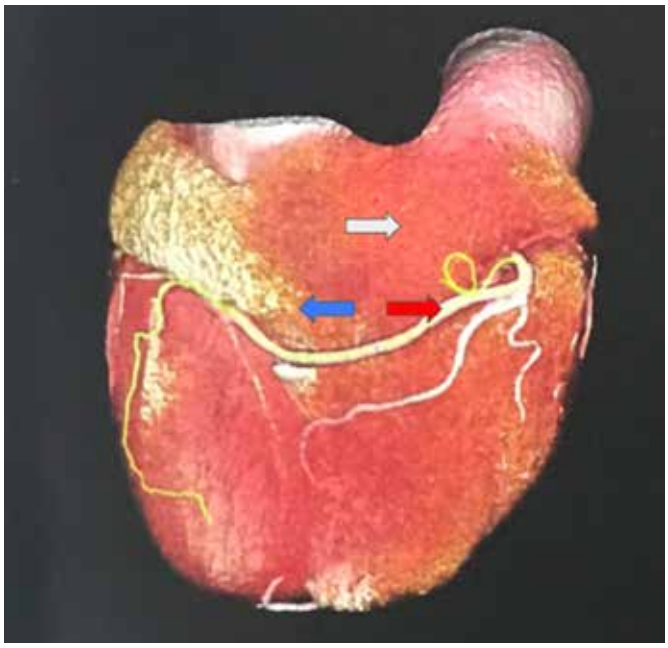

Figure 02: Three-dimensional representation from coronary angiotomography of the patient's heart in a right posterolateral view shows the enlarged coronary sinus (blue arrow) above the right coronary artery (red arrow) and right atrium (white arrow), with the right superior vena cava not visualized in this view.

DISCUSSION

PLSVC can be relatively common, as it is the most frequent among thoracic venous anomalies. While PLSVC is mostly isolated, its coexistence with ARSVC constitutes a rare congenital venous malformation [5]. In a normal adult, about one-third of venous return is through the superior vena cava, and in cases of obstruction, venous content will flow through collateral circulation to the lower part of the body, reaching the atrium through the inferior vena cava. This usually takes several weeks for collateral vessels to dilate sufficiently to accommodate the flow from the superior vena cava, leading to an increase in cervical venous pressure by 20 to 40 mmHg [6]. Difficulty in draining the superior vena cava can result in edema in the upper part of the body, potentially causing cerebral edema and airway edema [6]. AR-

SVC is closely related to PLSVC, as diverting drainage to the left allows venous return with less resistance and, therefore, with fewer or even no clinical repercussions. However, generally asymptomatic ARSVC with PLSVC can be a component of more complex cardiac pathologies or, in stressful situations, may lead to significant issues such as various arrhythmias [3].

PLSVC, in addition to the vascular changes already described, is often associated with other congenital heart lesions, especially when it drains into the left atrium [4]. Approximately 92% of PLSVC drain into the right atrium through the coronary sinus; however, the remaining 8% drain into the left atrium independently of the coronary sinus, creating a left-to-right atrial shunt [7]. Atrial septal communication has been the most common cardiac anomaly associated with the left superior vena cava [4]. Other cardiac lesions accompanying this anomalous vessel include single atrium, ventricular septal defect, Eisenmenger complex, Tetralogy of Fallot, truncus arteriosus, pulmonary stenosis, tricuspid atresia, aortic coarctation, anomalous pulmonary venous return, and absent right superior vena cava [4]. In cases diagnosed with Tetralogy of Fallot and Eisenmenger syndrome, there is a 20% and 8% probability of PLSVC, respectively [8]. There is also a frequent association with situs inversus or partial transposition of the viscera and/or levocardia [4]. A high incidence of left superior vena cava has been reported in asplenia [4].

In cases where right drainage is predominant, the coronary sinus typically shows an expansion that can lead to the compression of the atrioventricular node and its surroundings. Other possible complications resulting from the volumetric increase of the coronary sinus include compression of the left atrium with reduced cardiac output and complications associated with mitral valve surgery due to anatomical proximity [3].

In patients with PLSVC, complications related to central venous puncture, regardless of the technique used, result from the tortuous path crossing structures under mechanical stress and therefore include angina, hypotension, and perforation, which can progress to severe arrhythmias, cardiogenic shock, and cardiac tamponade. Another relevant risk described in the literature is stenosis of the coronary sinus, which can lead to the failure of coronary drainage with consequences for the arterial flow of the right and left coronary arteries [3]. In the specific case of pacemaker implantation, there are difficulties in fixing and positioning intracavitary electrodes.

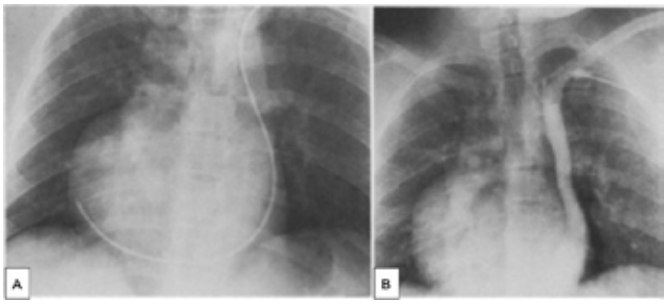

Figure 03: A: Shows that the catheter travels through the left paramediastinum, the left cardiac border in the left superior vena cava, and in the coronary sinus. B. Contrast infusion highlights the left superior vena cava and the coronary sinus[4].

CONCLUSION

PLSVC is an infrequent anatomical condition, so routine screening in preoperative evaluation is not necessary. Due to its predominantly asymptomatic nature, PLSVC may not be diagnosed until invasive procedures involving the vena cava occur. Although routine screening is not recommended, it is prudent to carefully observe exams that may show and facilitate early diagnosis, such as X-rays, computed tomography, echocardiography, and others. Thus, incidental findings, when diagnosed, should be communicated to the entire team, allowing for the proper prevention of associated complications. The anesthetist, as a participant in patient care, should be able to recognize and assist in preparing the team and managing the patient.

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LOW-GRADE MALIGNANT PHILOID TUMOR ASSOCIATED WITH DUCTAL CARCINOMA IN SITU: A CLINICAL CASE REPORT

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ABSTRACT

Phyllodes tumors are rare in the breast, ranging from benign to malignant, rarely associated with carcinomas, especially ductal carcinoma in situ.
OBJECTIVE: To describe a clinical case of low-grade malignant phyllodes tumor associated with ductal carcinoma in situ GN2.
CASE REPORT: Patient E.R.A., 53 years old, presented with a rapidly growing nodule in the left breast in the upper lateral quadrant. Physical examination revealed a well-defined, hardened, and painless nodule. Mammography showed a dense, well-defined, 20 cm nodular lesion in the same location. Following a Fine Needle Aspiration Biopsy (FNAB) with negative cytology and clinically negative axillary nodes, the patient underwent a quadrantectomy to remove the tumor with clear margins. Pathological evaluation confirmed a low-grade malignant phyllodes tumor associated with ductal carcinoma in situ, displaying moderate cellular atypia and high expansibility. The diagnosis was confirmed through immunohistochemical analysis, showing positivity for specific markers.
DISCUSSION: Phyllodes tumors (PT) are more common in the 4th to 5th decades, especially among Asian and Latina women. They are typically painless, large, firm, and rarely associated with carcinomas. Considerable growth (up to 41 cm) is common, and axillary involvement is rare. Diagnosis is challenging due to unclear imaging findings. Surgical resection with clear margins is the preferred treatment.
CONCLUSION: Epidemiological studies are essential for standardizing phyllodes tumor management, and due to the lack of data, complete resection with clear margins is the best approach.

KEYWORDS: NON-INFILTRATING INTRADUCTAL CARCINOMA; BREAST NEOPLASMS; PHYLLODES TUMOR

INTRODUCTION

Phyllodes tumors (PT) are a rare condition, accounting for 0.3% to 1% of breast neoplasms, making comprehensive studies on this lesion and its prognosis challenging. In histopathology, a fibroepithelial tissue with a foliaceous structural pattern is observed, featuring clefts lined with epithelial cells and hypercellularized stroma, justifying the name PT (ZHANG; KLEER, 2016). According to the World Health Organization (WHO), the malignant or benign nature is determined based on characteristics identified through microscopy, considering factors such as cellular features, stromal tissue cellularity, presence or absence of nuclear atypia, mitotic rates, among others (TAN et al, 2012). The histological features of the PT and fibroadenomas can overlap, with lobular duct involvement and stromal cellularity being compatible with PT (TAN et al., 2012). In the literature, it is recognized that malignant PT have a lower prevalence compared to benign tumors, and when malignancy is present, distant metastases occur in approximately 22% of patients (TAN et al., 2012; PAPAS et al., 2020).

In the worldwide literature, data on PT is scarce; however, it is known that the first report of malignancy occurred in 1931 after the identification of tumor metastasis to the lung (LEE; PACK, 1931). Thus, it can be observed that recurrences do not occur in the majority of patients, and abnormal growth, exceeding 10 cm, is not uncommon in these cases, referred to as giant PT (PAPAS et al., 2020). Epidemiologically, these tumors are more frequently diagnosed in patients with an average age of 45 years, with a predilection for the upper outer quadrant of the breast (PAPAS, et al., 2020). These aspects should be considered, as PT can be confused with fibroadenoma lesions, and the combination of histological and clinical factors can aid in the differential diagnosis (TAN et al., 2012).

The concomitant occurrence of Phyllodes Tumors (PT) with other lesions is uncommon and poorly reported, especially in carcinomas, and when present, in situ lesions show higher recurrence rates (OZZELLO; GUMP, 1985). The relationship between these two lesions is not clear, despite various existing theories attempting to provide a possible explanation. One of these theories suggests the

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sudden transformation of the epithelium into carcinoma cells or that the transformation of these cells occurs randomly (NISHIMURA et al., 1998). Additionally, the search for molecular markers that could correlate the two diseases does not contribute to explaining the potential associations (TSE et al., 2002).

The aim of the present study is to report a clinical case of a 20 cm low-grade malignant phyllodes tumor associated with ductal carcinoma in situ in a 53-year-old patient.

CASE REPORT

The patient, E. R. A., 53 years old, sought medical attention complaining of a painless, rapidly growing tumor mass in a large part of the left breast. On physical examination, a palpable nodule was noticed in the upper lateral quadrant (ULQ), with relatively well-defined borders, hardened consistency, and painless character. Additionally, no palpable lymph nodes were detected in the axillary or supraclavicular fossa. The mammography revealed a dense nodular image, rounded in shape, with well-defined contours, measuring 20 cm, located in ULQ (Figure 1).

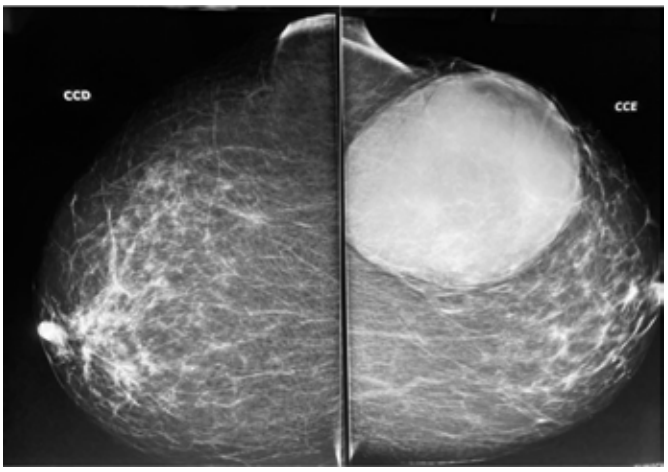

Figure 1 - Mammography revealing a well-defined border tumor mass in the left breast with a diameter of 20 cm.

Fine Needle Aspiration (FNA) was performed, and the cytology results were negative. Axillary staging was not detected. The patient was, therefore, subjected to surgical treatment through Quadrantectomy for tumor removal, with safety margins along the resection (Figure 2). An anatomopathological evaluation confirmed a lesion compatible with phyllodes tumor associated with low-grade ductal carcinoma in situ, moderate cellular atypias, and a high expansibility grade (Figure 3).

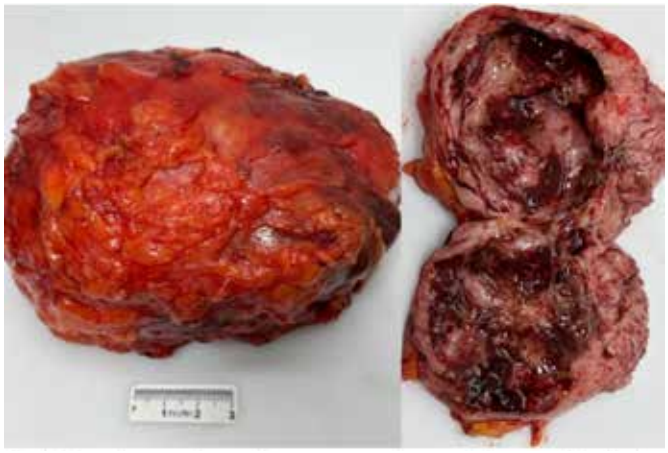

Figure 2 - Lesion removed with safety margins, showing the internal and external aspects, with brown-red coloration, solid, and well-defined.

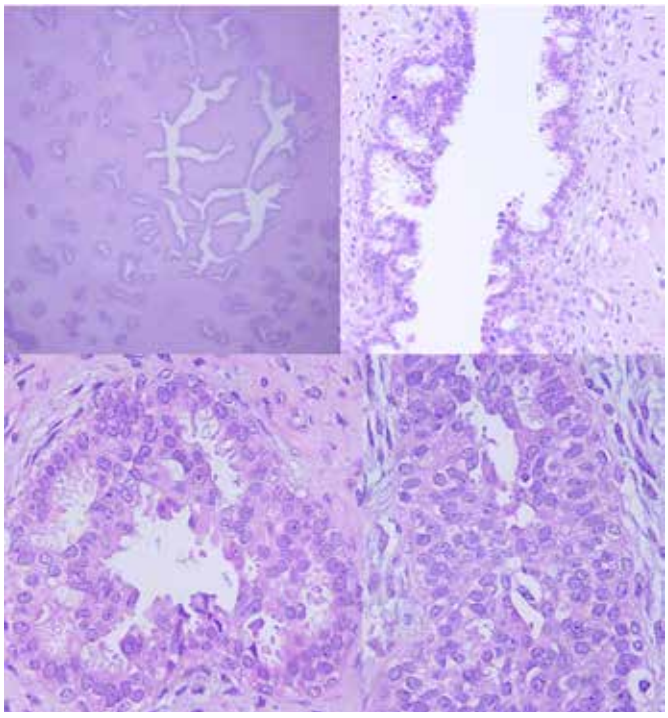

Figure 3 - Microscopic aspect of the TF lesion in the upper photos, observing the foliaceous appearance. In the lower photos, carcinoma in situ.

The diagnostic definition occurred with an immunohistochemical study that showed basal cell positivity for p63 protein, 5% expression in the portion affected by carcinoma in situ for the Ki-67 marker, and 10% for stromal cells, ER+, PR- (Figure 4).

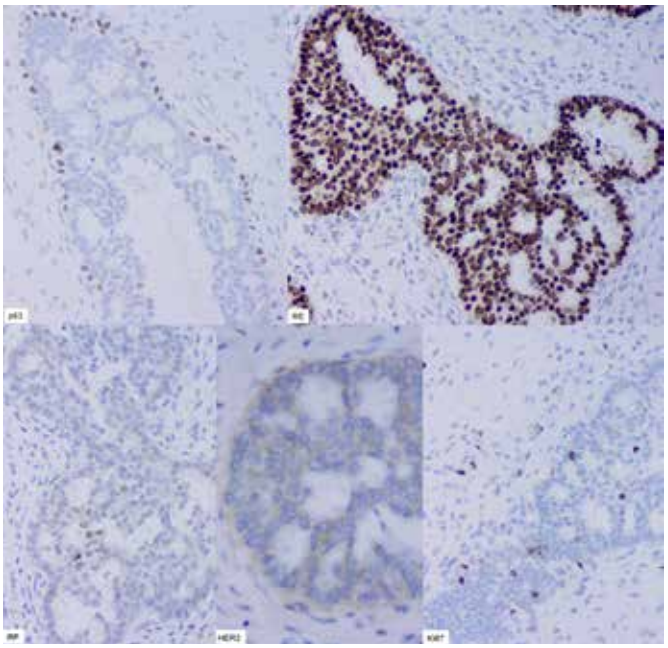

Figure 4 - Immunohistochemistry showing positivity for p63 protein, ER+, and Ki67.

DISCUSSION

The present study is characterized as a case report aiming to describe the diagnostic and therapeutic process for the treatment of a patient with Phyllodes Tumor (PT). According to worldwide literature, the occurrence of these lesions is more prevalent between the 4th and 5th decades of life (PAPAS et al., 2020). In a recent systematic literature review, the authors found that patients were typically between 35 to 55 years old (YU; HUANG; TAM, 2022). Additionally, it is known that palpable axillary lymphadenopathy is not routine in the presence of PT, and the preference for the upper lateral quadrant of the breast occurs in 35% of cases (GULLET; RIZZO; JOHNSTONE, 2009). In this study, the patient was diagnosed at the age of 53, with a tumor in the upper lateral quadrant of the breast and without lymph node involvement, characteristics that are consistent with the reported data.

The tumor's growth occurs in two phases, with the initial phase characterized by slow growth, and in advanced phases, the lesions commonly can reach 10 cm or more (TAN et al., 2012). Recent data indicate that the average size of tumors at the time of diagnosis can range from 2 to 10 cm (YU; HUANG; TAM, 2022). However, in other studies, authors identified that sizes can be extremely variable, ranging from 1 to 41 cm, with an average of 4 to 7 (FERNÁNDEZ-FERREIRA et al., 2021). Despite this, tumors larger than 10 cm in diameter represent a minority of cases, accounting for 10% of lesions (LIANG et al., 2008). Thus, the patient in the presented case corresponds to this minority group as her diagnosis occurred when the tumor measured 20 cm. This demonstrates how delayed the seeking of professional help was.

The use of imaging exams can be helpful in diagnosis; however, clinical and radiographic data should be combined, especially for differential diagnosis from fibroadenoma (FERNÁNDEZ-FERREIRA et al., 2021). Although there is a similarity in appearance, the large size and rapid growth are suggestive of PT. Additionally, mammography shows the tumor as a smooth, multilobulated mass, and on ultrasound, the lesions are hypoechoic, solid, with partially circumscribed indistinction (GULLET; RIZZO; JOHNSTONE, 2009).

Despite the limited existing data, it is known that surgical treatment with complete excision is still the best form of treatment, with safety margins greater than 1 cm recommended (FERNÁNDEZ-FERREIRA et al., 2021). Additionally, the use of radiotherapy is only necessary when applying safety margins is not possible, as the risk of recurrence increases in such cases (BARTH et al., 2009). The scarcity of data does not allow for definitive conclusions regarding the use of chemotherapy in PT, and its indication is still limited (FERNÁNDEZ-FERREIRA et al., 2021).

The association of PT with other lesions is rarely reported in the literature, with carcinoma in situ being the most frequent type of lesion (NOMURA et al., 2006). However, it is still unclear which factors may contribute to the concurrent occurrence of these lesions, and when it does occur, the secondary diagnosis often happens at the time of excision and histopathological analysis of the samples (NOMURA et al., 2006). Therefore, detailed handling of the tumor is essential, and, as in the present case, the evaluation of the association with carcinomas can be detected early.

CONCLUSION

Epidemiological studies should be conducted to assist in standardizing approaches related to the management of PT, especially in cases of associations with other types of tumors. Due to the scarcity of data, complete excision of the lesion with adequate margins is currently the best way to control this neoplasm.

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