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PHANTOM LIMB SYNDROME, INVOLVED NEUROLOGICAL MECHANISMS AND CASE DESCRIPTION IN AFFECTED PATIENT

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Abstract: A case of a 27-year-old male patient is described, in which at the age of 14 his life completely changed. While trying to catch a kite, the young man received an electrical shock that caused him to lose most of his legs and left arm. "During the electrical discharge, it felt like I was exploding, it was like my whole body was being pierced, I felt like I was cracking and the shock was detonating everything." He was hospitalized in serious condition, underwent more than 10 surgeries and remained in the hospital for more than two months until returning home, in Campos dos Goytacazes, in Rio de Janeiro. [1]. Currently, the patient still continues to experience stimuli in the amputated regions, such as pain, cold and heat, which in medicine is known as phantom limb syndrome.

Keywords: Phantom Limb Syndrome, Electrical Discharge Accident, Surgical Procedures.

Figure 1. Illustrative figure.

INTRODUCTION

Phantom Limb Syndrome is a neurophysiological condition in which the amputated part becomes detached from the body, but not from the brain, causing severe pain. [2]. Most people who have lost a limb can still feel it. These individuals can flex the lost areas or even feel the irritation of a bracelet on the lost arm, which suggests the existence of a kind of map of the organism well distributed in our brain. For a long time, the belief was held that sensations arising from missing segments of the body had a psychological origin, which changed from the 21st century onwards, when this belief was replaced by the "physiological theory". This change occurred due to advances in scientific research that linked phantom limb syndrome to areas of the post-central cortex, determining somatosensory maps [3]. While in Brazil there are no statistics on phantom limb pain syndrome, in the world literature up to 85% of amputee patients experience this painful entity. With several pathophysiological mechanisms involved in its origin, the most common forms of treatment are not very effective in controlling pain, making it necessary to use interventional techniques such as neuromodulation. [4]. This pathology can be extremely resistant to treatments and pose a unique challenge in managing pain in a limb that is no longer present. [5]. Given the lack of long-term efficacy in the drug management of Phantom Limb Syndrome in some patients, the search for alternatives must be a relevant concern for health professionals. [6]. Patients who undergo amputation procedures routinely complain of pain in the amputated limb. This type of pain is commonly referred to as phantom pain. Unlike stump pain, the sensation of phantom pain is considered illusory, as the amputee reports perceiving the presence of the missing limb as still active. Phantom pain usually presents in the first week, after separation of the amputated limb. However, it can appear after months or even years, with duration and symptoms that vary from case to case. It is worth noting that not everyone who has had a limb amputated feels this type of pain. The phantom sensation is so real that, in some cases, hand amputation patients have reported that they have tried to answer the phone with the missing hand. Other reports present situations in which patients with amputated legs tried to get

up to walk. There are several speculations about the possible causes of phantom pain. According to Moraes et al, there are three main mechanisms involved in phantom pain, they are: "peripheral, spinal and cerebral factors. Which are responsible for countless psychological, physical and environmental triggers."[7].

In Brazil, it is estimated that the incidence of amputations is 13.9 per 100,000 inhabitants/year (around 40,000 amputations/year), with lower limb amputations corresponding to 85% of all limb amputations [8]. The main indications for such amputations are divided into 2 large groups: 1) elderly group, due to complications from chronic degenerative diseases (up to 80% of these resulting from peripheral vascular diseases or diabetes). 2) young adult men, due to traumatic external causes (mainly traffic accidents and gunshot wounds). Meanwhile, at a global level there are controversies regarding the number of amputations, ranging from 2.8 to 43.9 per 100,000 inhabitants/year, depending on the country analyzed. [9]. Brazilian epidemiological data on Phantom Limb Syndrome are not yet known. Among the various reasons that must be taken into account for such variation, we can mention: type of population, study design and method of clinical evaluation of individuals [10]. This pathology can be extremely resistant to treatments and pose a unique challenge in managing pain in a limb that is no longer present. Considering the lack of long-term effectiveness in the drug management of Phantom Limb Syndrome in some patients, the search for alternatives must be a relevant concern for health professionals. [11].

The objective of this work is to describe the case of a young male, 27 years old, in whom, at the age of 14, he suffered an accident while trying to remove a kite that was stuck in the electrical voltage network, which caused him to suffer an electrical discharge so intense that it led to the burning of the muscular tissues of his two legs and his left arm, and this resulted in the amputation of these limbs by a surgical procedure, carried out by a doctor in the city of Rio de Janeiro, Rio de Janeiro, Brazil [12]. The young man was hospitalized in serious condition, underwent more than 10 surgeries and remained in the hospital for more than two months until returning home, in Campos dos Goytacazes, in Rio de Janeiro, Brazil [13].

The young man currently reports: "I can feel my entire left arm, which was the amputated arm, I can move all my fingers and with each movement I feel a lot of pain. In my right leg, which is up to mid-thigh, and in my left leg, which is up to the knee, I also feel a lot of pain. [14]. The pain I feel in my amputated limbs is not constant pain, but rather at certain times of the day."

This young man had his left arm amputated up to the muster, and surgery was performed on his right arm so that the patient could have mobility in that limb (Figure 2). In this surgery, the doctor removed a portion of the serratus anterior muscle (Figure 3) and inserted it into his right arm, seeking to reconstruct the limb.

Figure 2. Right arm of the patient in the case presented.

Figure 3. Patient in the case presented, where muscle fibers were removed.

MATERIALS AND METHODS

This research is a systematic search for a literature review, and presentation of a case in which the patient suffers from phantom limb syndrome [15]. The article selection process was carried out through three stages, namely: first stage, the search for articles in the databases using keywords. In the second stage, duplicate articles and those that strayed from the topic were excluded, observing the possible relationship with the objective of the study and, finally, the third stage, comprised the process of careful reading of the articles selected for this research. [16].

As exclusion criteria, literary materials that had no direct relationship with the proposed topic were rejected, such as case studies and articles whose themes were about phantom pain in body organs. [17]. Through this, the main advances in understanding the pathophysiology and progress in treatments for phantom pain were sought. After carrying out all the search steps and determining the articles that met the inclusion criteria and consequently were instruments of this review.

RESULTS

After analyzing the case of young MB, and through bibliographical reviews, it is possible to conclude that he suffers from phantom limb syndrome, due to his entire neurological mechanism that is still involved in the regions dismembered by the accident. As is the case in approximately 85% of amputation cases, whether due to trauma or surgical procedures, the region still continues to be stimulated, even in the absence of the limb.

DISCUSSION

The pathophysiology of Phantom Limb Syndrome is not yet fully known, and is still explained only by models based on animal studies that are already well accepted in the scientific community, however without evidence at statistically significant levels in human studies. [18]. Among the currently well-accepted explanatory models are morphological, physiological and chemical changes in both the central and peripheral nervous systems, being influenced by factors such as genetic predisposition, nature of neural damage, in addition to psycho-emotional and sociocultural factors. Peripheral mechanisms include: ectopic neuronal activity of afferent nerves originating from a neuroma in the amputation stump ("stump pain") and abnormal activity of axons of the dorsal root ganglia due to the activation of resistant sodium channel subtypes. to tetrodotoxin manifested in injured neurons [19]. Dorsal root ganglion cells begin to show abnormal activity in response to common stimuli or even spontaneous activity, becoming a source of afferent information to the posterior horn of the spinal cord. [20]. Such abnormal activities in dorsal root ganglion cells may be the result of an increased afferent barrage of signals to such cells, due to an increase in the expression of sodium channels. [21]. The first theories about the pathogenesis of Phantom

Limb Syndrome located its origin in the stump of the amputated limb, postulating that ectopic discharges from the neuroma were the main source of pain generation. [22]. Lidocaine injections into the amputated limb stump neuroma resulted in a reduction but not cessation of pain, suggesting that this is an incomplete explanation but that neuromas may evoke or increase painful sensations perceived in the amputated limbs. [23]. Among the central mechanisms we have: cortical reorganization and spinal cord sensitization [24]. Limb deafferentation induces reorganization in the somatosensory cortex [25]. Similarly, thalamic plasticity and reorganization of the motor cortex have been observed after amputations and may have relevance in the induction or maintenance of Phantom Limb Syndrome. [26]. One of the approaches most described in the literature was called "mirror therapy"."[27]. This is a technique applied using a mirror positioned in the sagittal plane, between the limbs to be approached, so that the mirror's reflection is aimed at the intact limb. [28]. When identifying the reflex of the healthy limb, amputee patients have a "virtual sensation" that their limb is amputated, which contributes to a neuromuscular and functional reduction [29]. In the management of phantom pain, it is believed that the therapy helps in better neuronal plasticity, with organization of somatosensory information in both afferent and efferent pathways. [30]. Mirror therapy was proposed by Ramachandran and Rogers-Ramachandran, and is based on the idea that the imaginary movement of the amputated limb could aid in the reorganization and integration between proprioception and neuromuscular functionality. [31]. In a randomized study carried out by Finn et al. researchers sought to improve phantom pain in the upper limbs using mirror therapy [32]. There was heterogeneity in the response to therapy, in general, for some patients the therapy was useful in reducing phantom pain, while for others there were no gains. [33]. However, in physical treatment situations with the mirror covered, pain worsened in some patients. [34]. In a theoretical model proposed by Rothgangel et al. researchers highlight the individuality of the response to mirror therapy [35]. The Therapy was tested remotely, using explanatory leaflets and DVDs, intense mirror therapy, including a session lasting up to 3 hours, and pre-recorded mirror therapy, in which the movements of the non-amputated limb were recorded on a mirror projection [36]. All interventions showed favorable results, with improvement in phantom pain in participants, however, the researchers emphasize the importance of an individualized approach, clear and precise guidance, in addition to the superiority of in-person guidance over remote guidance. [37]. Wareham and Sparkes (2020) state that up to 70% of military amputees suffer from phantom pain, which is difficult to manage, and which significantly compromises their quality of life and functionality. [38]. According to the study, phantom pain in the limbs is generally associated with cortical disorganization, associated with impaired laterality. [39]. The study did not find unanimous benefits of pain and laterality after ET session. However, among the 16 participants, two reported improvement in functional limitation and reduction in pain, respectively. [40]. Given such data, further studies with a greater number of sessions, as well as a more significant number of participants, are suggested. [41] Other non-pharmacological resources have also been reported to address phantom limb pain. [42]. In a clinical trial carried out with 21 patients with phantom pain refractory to other treatments, percutaneous image-guided nerve cryoablation was performed, with a substantial improvement in functionality and reduction in pain scores. [43]. Before the procedure, patients had a baseline mean pain

score of 6.2 with a subsequent reduction in pain levels to 2.0. [44]. Cryoablation or cryoneurolysis has historically been used to manage pain in metastatic diseases, although it is an innovative resource [45]. Phantom pain and its main therapeutic approaches: literature review phantom limb pain management is a consensus in the literature on the need for new studies, as well as the development of validated protocols for treatment [46]. Fifteen lower limb amputee patients who underwent acupuncture for a period of four weeks were included in the study. [47]. Fifteen lower limb amputee patients who underwent acupuncture for a period of four weeks were included in the study. [48]. There was an improvement in the average intensity of pain, up to one month after the acupuncture sessions, however, the study does not address the association of other techniques or treatments by users, which may also influence the results obtained and/or observed. [49].

CONCLUSION

Phantom pain is recurrent in individuals who have had a limb amputated. The pathophysiology of phantom pain is still not well understood, despite a good collection of research on the subject, there is great similarity between the articles, thus restricting the content covered. [50]. There is still no clear evidence or mechanism that explains why some people develop this phantom pain. Therefore, there is no specific treatment for this phenomenon. [51]. We therefore understand that the first step towards effective treatment is to recognize that the pathology exists and welcome the person who experiences this syndrome, without pre-judging the matter. [52]. The next step is the application of an integrated treatment involving the areas of psychology and physiotherapy, after all, the mind and body are interconnected. We concluded that the most applied treatments are mirror therapy, transcutaneous electrical nerve stimulation, hypnosis and biofeedback. [53]. It is important that the treatment is carried out by a multidisciplinary team. Phantom limb syndrome can be refractory to clinical treatment in some patients, causing great suffering. [54]. Spinal cord stimulation appears as a therapeutic option in selected patients [55]. It is a reversible, adjustable and minimally invasive neuromodulatory technique. It provides satisfactory results in the short/ medium term, but as demonstrated in the articles, sustained long-term results remain in a small number of patients. For this reason, strict criteria must be used when indicating [56]. Due to the presence of several limiting factors among the studies analyzed, new studies with standardized and optimized methodologies are needed to more effectively evaluate the factors involved in spinal cord stimulation for the treatment of phantom limb syndrome.

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