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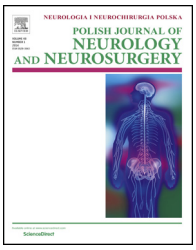

Review article

Phantom phenomena and body scheme after limb amputation: A literature review

Aneta Pirowska [a], *Tomasz Wloch* [b,c,*], *Roman Nowobilski* [d], *Maciej Plaszewski* [e], *Abdellah Hocini* [a], *Doménico Ménager* [a]

[a] *Institut Robert Merle d'Aubigné, Artificial Limb Centre (CRA – Centre de Reeducation et d'Appareillage), Valenton, France*
[b] *Department of Rehabilitation in Internal Diseases, University School of Physical Education, Krakow, Poland*
[c] *The University Hospital, Krakow, Poland*
[d] *Institute of Physiotherapy, Faculty of Health Sciences, Jagiellonian University Collegium Medicum on School of Medicine, Krakow, Poland*
[e] *Faculty of Physical Education and Sport in Biala Podlaska, University School of Physical Education, Warsaw, Poland*

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ABSTRACT

Phantom phenomena are subject of various, often inconsistent, descriptions, and new concepts and treatment approaches emerge. The aim of the study is to describe contemporary terminology and developments in the field, and to share personal experience.

A review of English and French language literature, published prior to 27th February, 2012, extracted from PubMed/MEDLINE, Google.fr, GoogleScholar databases, and by hand searching of selected full text papers and textbooks with correspondence to personal clinical experience was performed.

The terminology and classification of phantom phenomena sensations, relations between intensity and character of phantom pain to the etiology and level of amputations, as well as the influence of presence and intensity of pre-operative limb pain and post-operative stump pain on phantom phenomena are described. The benefits of mirror therapy and early introduction of prosthesis and applying functional prosthesis are also presented, with a glance at other conservative and surgical treatment approaches.

1. A brief historical overview

Historically, phantom phenomena were interpreted as a form of psychical or mental disorders [1,2], otherwise as a proof for existence of immortal soul[1] [3–5]. Admiral Horatio Nelson, who lost his arm during the attack at Santa Cruz de Tenerife on 25 July 1797, and as it is believed [6,7] (personal communication), described that in one of his letters. First, dated at 1551, dramatic description of feeling of an already lost limb was authored by Ambroise Paré (1510–1590) [8,9], famous French royal surgeon. Paré, performing amputations and applying prostheses, noticed the occurrence of phantom phenomena. He claimed that the phantom pain is a consequence of the stimulation of the nerves of the stump [10].

In 1872, an American army surgeon Silas Weir Mitchell published in a neurological journal *Injuries of Nerves and their Consequences* [9,11], an article *Neural maladies of stumps* and

* *Corresponding author at*: II Katedra Chorób Wewnętrznych UJ CM, ul. Skawińska 8, 31-066 Kraków, Poland.
E-mail address: twloch@wp.pl (T. Wloch).

[1] Nelson concluded the presence of his phantom limb as a '*direct evidence for the existence of the soul*' [2–4].

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introduced the term 'phantom limbs' in the official terminology [9,12].

2. Contemporary classifications of phantom phenomena

Currently, *phantom phenomena* are divided into categories of *phantom sensations* (pain-free reactions from the amputated extremity) and *phantom pain* (a pain from amputated extremity or from another amputated part of the body) [13,14]. Hunter et al. [15,16] introduces a term of *phantom awareness*, describing the consciousness of the presence of the lost part of the body ('a general awareness of the missing limb'), rather than the perceptual reactions referred to it.

Phantom pain is also defined as a frequent type of the feeling of existence of the missing extremity [17] or of the deafferented body part characterized by specific form, weight or range of motion [14,18].

According to Hunter et al. [16], the awareness of phantom is necessary for phantom pain to occur, not the opposite. In our concept of the paper, we decided to exclude the phenomenon of stump pain (i.e. the remaining part of the amputated limb) from the discussion, since, in our view, and in accordance to Finoff [1], this kind of sensation wanes relatively quickly, as the postoperative wound heals.

Phantom limb pains take a significant place among neuropathic pains [19–21], which, in turn, are qualified with inflammatory and dysfunctional pains, as chronic pains in the general qualification of pain [20,22,23]. The cause of chronic neuropathic pain is the injury of peripheral nervous system [24]. Genetic predisposition [24] and the influence of psychosocial factors [1,25,26] are also underlined.

Phantom phenomena are present in about 80% of amputees [20,27–31]. According to Merskey and Bogduk [23] and Haug [32], about 70% of the amputees will sooner or later experience phantom sensations. The phantom phenomena may occur immediately after the amputation or many months, even years later [14,29,31,33].

3. Diversity and variability of phantom phenomena

Intensity, duration and repeatability of the sensations are inconsistent and individually variable [14,16,31,34].

At times, the feeling of phantom is so strong and realistic, that the patient forgets that their amputated limb is shorter and tries to stand on it, falls and experiences injury [35]. Chodak [36] describes the impression of phantom handgrip, painful sensation of fingers digging into his phantom palm[2] [37], freezing of the hand, involuntary, rigid flexed position of the leg, with parallel realistic sensations, such as: feeling of warmth, sweating and weight. Phantom phenomena are not limited to extremities and may also follow amputations of other parts of the body [13,23,38].

Blumenthal [39] reports that the prevalence of phantom pain in the first six postoperative months is estimated at 50–75%, and gradually diminishes during subsequent two years. The majority of patients (85–97%) suffered from the phantom pain during first month, while the problem was present in 60% of cases after a year post operation, when only 10% of the studied amputees reported this kind of pain for the first time. In a study by Casale [2] phantom pain and phantom sensations were reported by 72% and 84% of subjects, respectively. Six months after surgery, the pain decreased in 67% of cases, while phantom sensations augmented in 90% subjects. Phantom pain was still existent in 60% subjects after a year and two years post surgery. Schley et al. [29] report, that in a studied cohort phantom pain and painless phantom sensations aggravated only in some subjects, and were experienced from several times a day to several times a year.

The intensity of phantom phenomena usually increases at rest, in the evenings and at night [40]. In a study by Kern et al. [41], of 537 amputees, almost 15% experienced no pain, 75% experienced phantom pains, 45% had stump pains, and about 35% perceived both. Over 60% of the amputees signalized sleeping disorders, in majority of cases connected with the phantom pain.

4. Phantom phenomena and the etiology of amputation

Some authors pointed to the correlations between the existence of phantom pains and the etiology of amputation [14,16,42], and others formulate contradictory opinions [43]. Hunter et al. [15,16] observed in a long-term study regarding vascular amputees, who were examined after 6 months, and between 1 and 3 years post surgery, that quality and quantity of the phantom sensations and phantom pain altered with time.

The dominance of phantom sensations and the frequency of their manifestation may be the most apparent within 3 months after upper or lower limb amputation. In lower limb amputees, at least two years post surgery, the frequency of the phantom pain and phantom sensations decreased, while the characteristics remained unchanged [16]. Similarly, according to Jensen et al. [27], phantom disorders are most intensive during first 6 months after surgery, and cease within 2 years.

To the contrary, Sumitani et al. [44] claim that majority of subjects experience phantom pains following several years post amputation. Desmond and MacLachlan [45] observed in their 50-year follow-up study a surprising figure of 42% and 43% of amputees still experiencing phantom and stump pains, respectively.

5. Mechanisms of phantom phenomena

The peripheral nerve involvement [18,31,44,46], the spinal cord [18,31,44,46] and the cortex [18,31,44,46] theories have been proposed to describe mechanisms of phantom phenomena. However, according to Richardson [18], the theories are in fact

[2] Similarly to the above mentioned admiral Nelson's case, who after amputation continued to be aware of the missing limb, in particular the existence of the fingers digging into the palm of his missing hand [4].

similar and correspond to the phenomena of the cortical reorganization and the pain memory.

The peripheral nerve involvement theory refers to inflammatory processes and neuromata of the cut nerve fibers of the stump [14,18,31,44,46]. As a consequence of amputation and deafferentation, pathological processes in the dorsal horn of the spinal cord may develop, and, consequently, cortical mechanisms leading to phantom pains may evoke [14,31,44,46,47]. Paradoxically, the limitation of this theory is the absence of phantom pains in some patients [14,18].

Thus, the investigations for mechanisms and causes of phantom pains are leading to the brain [14,31,46]. In 1993, Robert Melzack formulated the *neuromatrix theory*, describing a genetically defined *neurosignature*, modified by the sensory input. The inputs have an effect on the neurosignature, which generates the output, in this case the post-amputation phantom pain [33,46,48,49].

Another phenomena is the reorganization of the *somatotopic map* – the representation of a part of body in the primary somatosensory cortex (S1) and the sensorimotor cortex [44,46,48,50], occurring after spinal cord injuries, the post-brachial plexus injury, in the complex regional pain syndrome (CRPS), and post amputation. For instance, phantom sensations from amputated limb may radiate to a particular part of the amputee's face [44,50].

A phenomenon of the *mirror visual feedback* (MVF) from the opposite hemisphere, leading to the modification of body awareness in the amputee, has also been described [51–53].

6. Phantom phenomena and body scheme

Self perception and perceiving of the amputees' bodies are a subject of various, often inconsistent, descriptions [43,54]. André et al. [43] proposed a general 4-category classification of forms of body scheme and phantom awareness (Table 1).

Initially, the phantom limb has a 'normal' size, and can subsequently remain unchanged or gradually disappear. A phenomenon of the telescoping may also take place, i.e. the distal part of the phantom progresses toward its proximal portion (the phenomenon of disappearing, telescoping of the limb) [14,18,55]. The telescoping may also regard to individual segments of the phantom, perceived as too short or too long, or may be manifested by the unperceiving of some of them [43,46]. Feeling of a foot directly 'implanted' into the knee joint or a *set of miniature toes*, 'joined' to the bottom of the stump, can serve as examples [18,56] (Fig. 1). Such a type of a deformed phantom is usually a subject of a long process of transformation, starting from the time of amputation [43]. Telescoping is less frequent in patients who suffered from a mechanical damage (e.g. due to injury) of peripheral nerves prior to amputation [57].

Table 1 – Classifications of body scheme perception in amputees (based on André et al. [43]).

Category	Description
I	Normal phantom limbs
II	Deformed or memorized phantom limbs
III	Illusion of the normality of the body
IV	Perception of the actual shape of the body

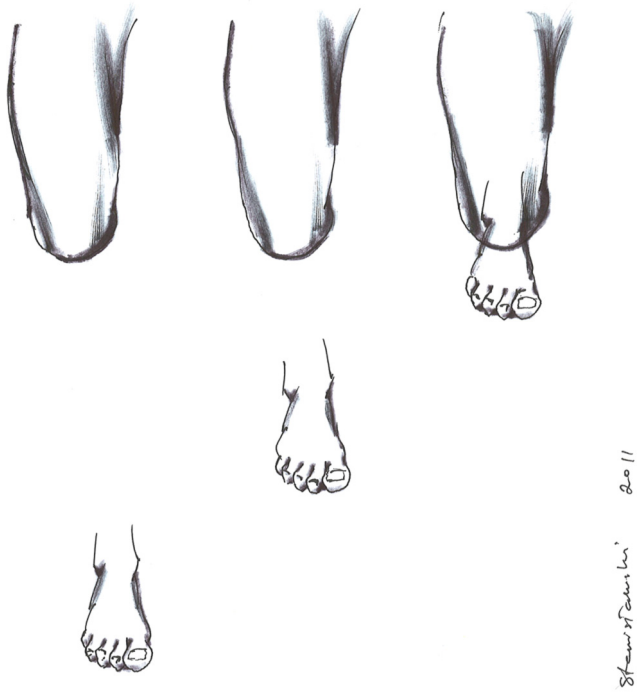

Fig. 1 – An example of so-called telescoping. Primarily, the phantom limb has a 'normal' size, and can remain unchanged in subsequent stages, gradually withdraw or may start to abbreviate in a telescopic manner, i.e. the distal portion of the phantom approaches the proximal one. The figure illustrates an example of a phantom foot awareness, without the awareness of lower leg, and an example of experiencing a foot directly ''implanted'' to the knee joint. Awareness of the lower leg may occasionally occur after the prosthesis had been applied.

According to Giummarra and Moseley [46], the phenomenon of telescoping occurs in about 20% of amputees. Occasionally, the telescoping may be composed into the sensation of *multiple phantoms* [58]. Lacroix et al. [59] described a case of a 16-year-old lower extremity amputee, who experienced three phantom limbs. A patient with a congenital deformity of her right leg, 10 cm shorter than the left one, at the age of 6 had undergone lower leg amputation of the right extremity. Ten years post operation the patient became aware of the three phantom limbs: a third, additional limb, as an imagination of the preoperative extremity, a second – phantom foot, telescoping directly from the stump, and a third, completing the amputated extremity, delivering the perception of the existence of a whole limb. Another case is the *multiple phantoms*, associated with the phenomenon of *a set of miniature toes* [58]. A cases of experiencing a second phantom limb in a shape of a set of miniature toes, attached to the distal portion of the stump, have been illustrated. Those toes were exceptionally delicate, continuously leading to a feeling of tingling and twisting [56,59].

The popular notion that the phenomena of telescoping reduces severity of phantom pains, has not been fully supported, and some authors separate these experiences [18].

Lower limb phantom phenomena are typically felt in toes, calcaneal tuber, metatarsus, or the talocrural and talocalcaneonavicular joints of the nonexistent leg [2]. They may completely or partially 'mimic' the lost part (e.g. a part of the foot) [56].

A position of the phantom extremity is usually identical to the position prior to the amputation, immediately before the nerve section during the operation or at the site of the injury. That fact is often found in traumatic amputations [60].

Typically, a phantom extremity acts as normal, in a natural position, e.g. remains flexed in the knee joint in a sitting position or moves depending on the movements and actions taken by the amputee. Ramachandran [61] describes an interesting case of a person with bilateral aplasic phantom upper limbs, experiencing phantom illusions, making gestures with their phantom limbs while conversing [58]. A phantom limb may also be perceived as unnaturally positioned, imposing inadequate behaviors (e.g. withdrawing wheelchair from a lift to avoid trapping of extended limb with the door, or moving sideward through the door due to the perceiving of phantom, adducted arm) (Fig. 2).

The phantom limb may even 'feel' moist, as stepping with the phantom leg in a puddle [62].

Sensations experienced as phantom limb movements may be volitional, controlled, otherwise unintentional – spontaneous or automatic, occurring mainly in young persons [54] (Fig. 3). Some amputees describe the embodiment and awareness of the phantom, without having any sensations from the lost part of the body [18].

7. Phantom pain in children and adolescents

Phantom pains are generally not reported in children below 4 years of age. Muller [54] claims that phantom phenomena in such young children are rare and temporary. Melzack et al. [63] studied 125 adolescents (mean age 14.7 years, no standard deviation stated), and found phantom phenomena to be present in 41 subjects, of whom 15 were characterized by congenital limb deficiency, and 26 had their limbs amputated before the age of 6. Similarly, Smith and Thompson [60] found phantom pains in almost 45% of 75 studied children. Finoff [1] concludes, that congenital limb deficiency leads to less apparent phantom sensations in comparison to children who had undergone amputations. Wilkins et al. [64] state that phantom phenomena appear mainly in those children, who experienced traumatic amputation, who manifest phantom sensations and phantom pains (100% and 83–100%, respectively).

However, in our practice we hardly ever notice phantom sensations and pains in children. An exception was a case of an adolescent girl who had had both legs amputated due to their congenital ectromelia and who experienced dreams in which she experienced awareness of both extremities.

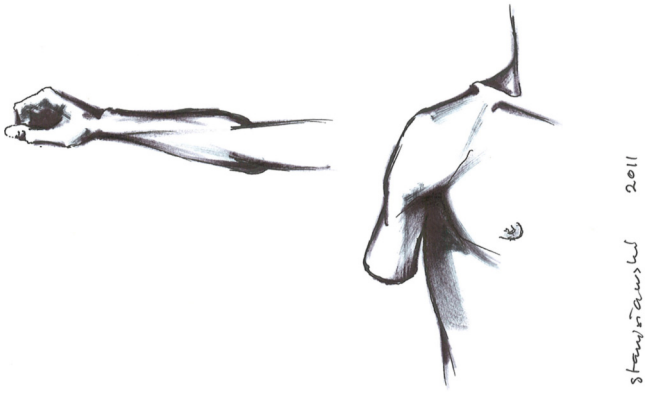

Fig. 2 – The phantom limb maintains an unnatural position, inducing inadequate behaviors of the amputee (e.g. passing sideward through open doors due to the awareness of extended phantom arm).

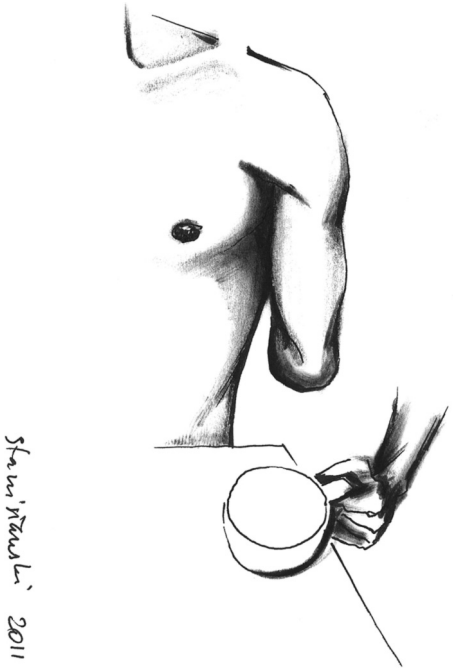

Fig. 3 – Amputees frequently experience a sensation of holding objects in their amputated hand. The attempt to collect them leads to unpleasant impressions, e.g. of pulling a cup out from the hand. Then, their phantom hand grasps the given object.

8. Mirror therapy, body scheme and phantom pain

A method referred to as the *mirror therapy*, is aimed to produce an illusion of a whole body scheme, despite the limb loss, and thus to decrease or suspend phantom pain. The method was introduced in 1996 by Ramachandran, initially for upper limb amputees, and subsequently for lower limb amputees [65–68]. A patient, when observing their existing extremity in a mirror or a mirror box, 'tricks' their mind and introduces false message on the existence of the amputated limb, thus diminishing pain symptoms. It can be even claimed that such message gradually 'amputates phantom pain' [66], which is in accordance with the notion of the nature of phantom pains,

originating in the central nervous system, not at the amputation site [62].

Application of mirror therapy also facilitates manipulation of the phantom limb, reducing its frequently unnatural alignment, or releasing its 'frozen' position [46,69]. Giummarra and Moseley claim that additional 'reduction' of the size of the reflecting limb with a lens improves the analgetic effect of the mirror therapy [46]. The method is reported to be effective, which we also observe in our practice, nonetheless its use is limited to single extremity amputees. Mirror therapy does not, however, prove to be effective in all cases of phantom sensations, which may be due to their complexity, as a phantom limb may be deformed, rotated or fallen to pieces, longer or shorter than the existing one [69]. As a result, the mirrored existing limb differs from the phantom picture/imagination of the lost extremity.

Seidel et al. [50] studied with the functional magnetic resonance imaging the activity of brain cortex in lower limb amputees participating in mirror therapy, and found that prefrontal cortical activity increased during the sessions. The authors associated this phenomenon and analgetic effects observed in the patients after completing twelve sessions of the therapy [50].

Moseley [70] and Moseley et al. [71] accent the effectiveness of the *graded motor imaginary therapy* (GMI therapy), consisting of the performing limb laterality recognition (utilizing photographs of the limbs), visualization (imagination) of the lost limb, and the mirror therapy [33,53].

Also, the *virtual mirror therapy* method, applying three-dimensional imaging, has been introduced [46,51,69,72].

In this case, the technique of *immersive virtual reality* (IMV) is used. The patient is 'immersed' in virtual reality, and begins to percept a virtual, complete body, which eventually leads phantom pain to deteriorate [37,46,68].

Sumitani et al. [44] underline the importance of the neurorehabilitative techniques for phantom pains, such as the *rehabilitation robot suit system*. Surgical procedures and spinal cord stimulation techniques are also utilized [73].

9. Phantom pain and preoperative pain

A number of authors underline the importance of the association between the presence of the preoperative pain with the severity of the post amputation phantom pain [29]. Dijkstra et al. [74] are more precise with the description of that relationship, claiming that the presence of the preoperative pain is connected with the risk of emerging of the immediate postoperative phantom pain. The authors claim that major risk factors of phantom pain are: vascular etiology of amputation, lower limb amputation, high level amputation, bilateral limb amputation, presence of phantom sensations, stump pain and the age at the time of amputation. Roullet and coauthors [75] confirm the theory of vascular etiology of amputation as a favorable factor of phantom pain. They also state that the intensity of immediate postoperative phantom pain depends on the strength of preoperative pain. According to Hanley and colleagues [76], the preoperative pain is a single predictor of the presence of chronic phantom pain two years post operation, and the existence of acute phantom pain is the only predictive factor of chronic phantom pain experienced after 6 months and after one year post amputation.

Also, regardless the etiology of amputation, cases of post-amputation pains, identical to those occurring prior to amputation (e.g. in-growing toenails), have also been documented [18].

10. Phantom pain and stump pain

The correlation between stump pain and phantom pains and phantom painless sensations has been evidenced [18,33,75]. Local anesthesia of the stump may temporarily moderate, whereas imposing external force (e.g. hit) on the stump, may enforce or increase phantom phenomena [57]. Patients who were subjected to epidural anesthesia and peripheral nerve block are less pronounced to phantom and stump pains in the first week post surgery in comparison to those in whom general anesthesia and spinal anesthesia were applied [14,77]. The differences were no longer observed 14 to 17 months post surgery [77]. The use of pre-emptive analgesia to protect from chronic pain after amputation due to critical ischemia of peripheral vascular disease has not been found effective [78].

According to Schley et al. [29], the probability or phantom phenomena to occur is higher in persons suffering from the stump pain.

11. Phantom pain and the level of amputation

Schley et al. [29] also signify the importance of the relationship between the level of amputation and the presence and severity of phantom pains. The authors observed that the majority of amputees who never experienced phantom pain, lost merely a part of a single toe, which supports the notion that above knee amputations are associated with more frequent phantom pains, in opposite to amputations below knee [79]. However, according to Nikolajsen and Jensen [42], emerging of phantom pain is independent from the level of amputation and side of the body. A more frequent occurrence of phantom pains after upper limb amputations, in comparison to lower limb amputations, has also been reported [14].

12. Multiplicity of phantom pains

Phantom pains are characterized by the variety of impressions, as it is described in Table 2.

Blumenthal [39] suggests that phantom pain is frequently accompanied by a feeling of a dislocated phantom limb, positioned divergently to the existing extremity. Aydin et al. [80] and Knarr [81] characterize phantom pain as an impression of spasm, piercing, twisting, dull aching, burning, and tingling. Vetrugno et al. [40] illustrate an incident of acute phantom pain, as a suddenly emerging feeling of tingling, burning, spasm, piercing, and squeezing. Though, sensations described by Muller [54] had a form of an electric shock.

Ramachandran and Altschuler [82] report that many patients can perform 'movements' with the phantom limb, and also may feel it as paralyzed, as 'cemented' or 'frozen in a

Table 2 – Characteristics of phantom pains.

Source	Most frequent characteristics of phantom pains
Blumenthal [39]	Burning, pricking, piercing, cutting, constricting, spasm
Aydin et al. [80]	Spasm, piercing, twisting, dull aching, burning, twinging
Knarr [81]	Spasm, burning, pricking with a knife
Vetrugno et al. [40]	Tingling, burning, spasm, piercing, squeezing
Muller [54]	Burning, spasm, tension, electric shock
Casale [2]	Sensation of heat, cold, itching, tingling
Ramachandran and Altschuler [82]	Sensations of: ''paralyze'', ''cementing of the limb'', ''freezing in an ice block''
Merskey and Bogduk [23]	Dull, burning, cramping pain, ''electric shock''
Sherman [85], Sherman and Barja [86]	Shooting pain, shaking, cramping, piercing, burning, squeezing
Wilkins et al. [64]; Sherman [85], Finoff [1]	Preoperative-like pain
Giummarra et al. [55]	Sensation of temperature, pressure, itching, vibrations, pricks

block of ice', and that phantom pain can be also intermittent or continuous, often with temporal severe exacerbations, which may lead to depression or even suicidal ideas. Phantom pain may be aggravated by emotional distress, cold, local irritation, orgasm in females and the post-orgasm period in males, bladder catheterization, defecation, micturition and smoking. An imitation of phantom pain by referred pain has also been reported. Heartache can refer to amputated left arm, and causal treatment of the referred pain may diminish or retain complaints [1].

13. Facilitating factors

Phantom pain can be triggered by either physical or psychological stimuli [31]. For example, a pain caused by an inflammation of the stump can provoke or exacerbate an already existing phantom pain due to personal or professional negative events. Other reported facilitating factors are: bilateral amputations, peripheral vascular diseases, diabetes, infections, gangrene, amputation of a distal part of a limb, presence of phantom sensations, late application, occasional or temporary use of a prosthesis [18,33], distress at work [33] or simply bad weather conditions [31].

Distress, anxiety and depression are also often in correlation with an augmentation of phantom pains [31,33]. Depression was diagnosed in 20–60% of amputees, a figure three to five times greater than in general population [33].

14. Phantom phenomena and prosthesis

Casale and coauthors [2] claim that daily use of prosthesis influences neither stump pain, nor phantom sensations, but definitely reduces phantom pain. The authors recommend wearing prosthesis for at least 9 h a day, but indicate that in case of upper limb amputations the influence of prostheses on phantom phenomena is more noticeable when using functional prostheses, in contrast to cosmetic prostheses. Using a functional or myoelectric prostheses by upper-limb amputees can increase the magnitude of phantom phenomena and reduce phantom pain [46]. Curelli et al. [26] found that phantom pains are more frequently reported by amputees in whom prosthesis has not yet been introduced.

Basing on André et al. [43], wearing and using of prosthesis decrease frequency of phantom sensations. Accordingly, Kauzlarič et al. [83] claim that the frequency of phantom pains diminishes proportionally to the duration of the reeducation with prosthesis. Merskey and Bogduk [23] find early introducing and using of the prosthesis important. In contrast, according to Hunter et al. [16], applying prosthesis does not show to be correlated with phantom pain depletion, and the fact of using functional or cosmetic prosthesis does not influence the intensity of phantom pains.

Our long experience confirms other authors' observations of beneficial influences of the prosthesis use on the diminution of phantom sensations. Still, appropriate fitting of the socket and proper, regular bandaging of the stump during the first year post amputation, are crucial. In opposition, improper fitting of the prosthesis leads to the alleviation or provocation of phantom pains.

When phantom pain appear, following the donning of a prosthesis, it is a very strong argument to suspect poor fitting.

Mayer et al. [84] observed in persons who had prostheses applied, that the scheme of their bodies did not alter, whereas in persons without prostheses phantom limbs became shorter (i.e. telescoping occurred) in a period of time extending six years after amputation. Applying prosthesis helps to maintain advantageous body scheme, in which phantom limb mimics residual one. However, applying prosthesis is not the most important factor influencing body image in amputees [84]. It is important, whether the prosthesis is seen as a part of the body, or an external object – in that case phantom pain is more feasible [81]. When applying prosthesis, phantom limb 'adjusts' to the prosthesis, like a hand wearing a glove [62].

Conflict of interest

None declared.

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Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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