Phantom Erection after Amputation of Penis. Case Description and Review of the Relevant Literature on Phantoms

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ABSTRACT: ***Background:*** Perception of a phantom limb is frequent after an amputation of an upper or lower extremity. Phantom penis is reported infrequently. ***Method:*** Case description and literature review. ***Result:*** The phenomenon of phantom penis followed total penectomy. Several aspects were unusual, particularly the existence with phantom only in the erect state, and associated recrudescence of a preoperative painful ulcer. General features of limb phantoms after amputation are reviewed including a résumé of recent studies of cortical reorganization. The phantom process is analyzed looking for clues to the nature of the underlying neural organization. The puzzle of phantom pain is briefly touched on. ***Conclusion:*** The development of the phantom is attributed to activity in the deafferented parietal sensory cortex.

RÉSUMÉ: Érection fantôme après amputation du pénis. Présentation de cas et revue de la littérature pertinente sur les fantômes. ***Introduction:*** La perception d'un membre fantôme est fréquente après une amputation d'un membre supérieur ou inférieur. On a rarement rapporté un pénis fantôme. ***Méthode:*** Présentation de cas et revue de littérature. ***Résultat:*** Le phénomène du pénis fantôme s'est manifesté à la suite d'une pénicectomie totale. Plusieurs aspects étaient inusités, particulièrement l'existence du fantôme seulement en état d'érection et associé à une recrudescence d'un ulcère douloureux préopératoire. Nous revoyons les caractéristiques générales des membres fantômes après amputation, incluant un résumé des études récentes sur l'organisation corticale. Le processus du fantôme est analysé pour chercher des indices sous-jacents sur la nature de l'organisation neurale. Nous faisons allusion à l'énigme de la douleur fantôme. ***Conclusion:*** Le développement du fantôme est attribué à l'activité du cortex pariétal sensitif désafférentié.

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The presence of a phantom limb is experienced by almost all amputees after amputation of an upper or lower extremity, and can be considered a normal physiological event.[1-3] A phantom breast after mastectomy is also well known.[4] Phantom penis is reported infrequently and only three references were found. A brief review of the literature at this point will provide the reader with some perspective on this rather esoteric subject. Afterwards the present case will be described.

Weir Mitchell in his original monograph – "Injuries of Nerves and their Consequences" included a foot-note reference to an unpublished case described to him by Ruschenberger of the U.S. Navy, of a penile phantom which was subject to erections.[5] Price Heusner's case 1[6] was a man aged 70 years who began to have intermittent erections two years after amputation of his penis. Prior to the amputation he had been impotent and lacked desire. The erection was not provoked by sexual phantasies. The phantom was so natural the subject was led to check for its presence visually. This state was brought to an end four years later when the subject suffered a gunshot wound of the spine productive of a paraplegia with loss of sensation for pain and temperature below the level of the navel. In Heusner's case 2 there was a painful phantom penis, not subject to erection.

Crone-Münzebrock[7] found in a follow-up study of 12 cases of penile amputation for carcinoma that seven experienced penile phantoms which faithfully reproduced the original member in size and position. All of these patients had residual penis stumps, 2 to 3.5 cm in length and voided via the stump-urethra. The passage of urine and any accompanying dysuria were felt in the phantom. Six of the seven retained their customary libido with normal erection and ejaculation. In two instances the phantom sensation had gradually been lost and was present only in the erect state. Two of the seven had transient pain in the phantom. The five cases without a phantom had lost their libido before operation.

The present case is of interest insofar as the amputation was total and the erect phantom regularly included recrudescence of a preoperative malignant ulceration with its accompanying pain.

Recent studies of phantom phenomena including pain, are providing new insights into the functional organization of cerebral cortical processes[8] and it is conceivable that cases like the present one could contribute to knowledge of one of nature's most basic functions.

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Case Report

A successful businessman, aged 44 years, developed a painful sore about 8 mm in diameter on the glans penis. Biopsy revealed carcinoma and he underwent penile amputation along with radical dissection of the inguinal lymph nodes bilaterally. One node on the left side showed the presence of carcinoma and maximum roentgen ray therapy was delivered. The amputation was total, that is there was no stump projecting anterior to the pubis. The patient voided via a perineal urethrostomy. The testicles were retained and bladder control was preserved. There was no recurrence of the malignancy.

At the age of 64 years, 20 years later, while under care for cerebral transient ischemic attacks the patient casually mentioned that since the operation and despite the absence of his male organ, phantom erections regularly occurred especially with erotic stimulation, for example, "seeing a pretty young woman". The phantom seemed to be of normal size, configuration and alignment, and was accompanied by a normal sexual feeling. So real was the experience that even after 20 years, the subject was still periodically obliged to check on the situation, tactually and visually. Particularly surprising to him was the exact reproduction on each occasion of the original painful sore, at the same site on the glans, accompanied by the same type and severity of pain as before the operation. The pain was apparently not severe. Restrictive undergarments were not an impediment. The patient was unaware of a phantom in the non-erect state. He gave no hint of having consulted the medical literature and he was not asked if he had discussed the matter with other physicians.

The patient died from prostatic carcinoma seven years later, aged 71 years, without further history or follow-up information. There was no postmortem examination.

Comment The history as recorded is probably reliable since it was proffered unsolicitedly. In retrospect many further details could have been sought that would have permitted a better comparison with what is known about phantoms at other sites, for example, the duration of the ulceration and pain before surgery, when in relation to surgery the phantom first appeared, the character and severity of the pain, changes in the phantom and pain with the passage of time, the duration of the phantom erection each time, an account of all the stimuli that were effective in eliciting a response, an analysis of the genito-pelvic sexual sensation, etc.

Discussion

A Brief Summary of Current Views on a Phantom Limb Phenomena. The special features of the present case will be better appreciated within the context of some knowledge of phantoms in general.[1-3] Phantom limbs are a normal consequence of amputation of extremities. Phantoms generally appear in the first few days after surgery and many patients, upon recovering from the anesthesia, feel that the amputated part is still present in its usual place. The surface contour of the three-dimensional limb phantom consists of a faint tingling feeling. Distal parts of a limb are plainer: hand and foot, and thumb, index finger and hallux, the regions with relatively larger cortical sensory representation.[9] The phantom is a purely somatosensory creation and can occur in the blind.

Amputees can usually move the fingers or toes but to a limited extent, and flexion better than extension. With changes of body position the phantom maintains a natural relationship to the stump. Painless phantoms especially of the arm undergo gradual telescoping in which the arm and forearm gradually fade from awareness and a normal-sized remnant of hand and fingers approaches or even penetrates the stump.

Pain of variable severity, from bothersome to highly distressing, may involve the phantom or stump or both. Pain in a phantom is usually a duplicate of pre-amputation limb pain.[10] A phantom part may bear an exact replica of an article worn preoperatively, ring, watch, shoe, bandage, etc.

Phantoms also occur after brachial plexus avulsion,[11] and after spinal cord transection with paraplegia.[12,13]

Regarding the neural mechanism underlying a phantom limb, the dominant view is that it is created by activity in the parietal sensory cortex that normally subserved somatosensation in the amputated part.[8] A sensation of tingling which reflects a partial deficit in the touch system at any level from the periphery to the parietal lobe, takes on the natural form of its amputated source. It might be expected that a preamputation painful neural pattern could co-exist. Older theories that attributed phantoms to activity in the severed nerves in the stump, or to psychological factors, have been discredited.[3] Surgical procedures which interrupt nerves peripherally, or tracts in the spinal cord or brain stem, fail to relieve painful phantoms.[14] Several reports describe relief from phantom pain, at least temporarily, as a result of surgical corticectomy, stroke or tumor involving the appropriate parietal cortex.[15-19]

Further evidence for cortical participation derives from studies of cortical sensory reorganization in amputees, based on the original work of Merzenich et al.[20] Pons et al.[21] found extension of facial responses far into the cortical arm area of primates with long-term deafferentation of an upper limb. Ramachandran et al.[22] in studying two subjects with arm-amputations, found that touch sensations on the face and on the stump, were referred somatotopically to particular regions of the phantom. The appearance of the phenomenon four weeks after the amputation was regarded as evidence against sprouting as the basis of reorganization, and in favor of the unmasking of preexisting silent connections. Kew et al.[23] used positron emission tomography to study the reorganization of cortical patterns in two patients with a deafferented amputated upper limb. Vibrotactile stimulation of the pectoral region on the involved side was referred to the phantom. At the same time blood flow was significantly increased in the corresponding hand-arm area of the cortex, representing an abnormal extension of 20 mm and l2 mm respectively, compared with the normal side.

In further experiments Ramachandran et al.[24] and Ramachandran and Rogers-Ramachandran[25] using a mirror-box, enabled subjects to move their immobile phantom fingers when the phantom was superimposed, visually, on the mirror image of their own normal moving hand. Also using a mirror-box, sensory stimuli delivered to the subject's normal hand were felt in the same place in the phantom. These results suggested that there is a considerable amount of latent plasticity in the adult human brain with pathways bridging the two hemispheres, emerging in less than three weeks. Chen et al.[26] using transcranial magnetic stimulation found that motor reorganization following amputation, occurs predominantly at supraspinal levels. Two investigators studied the relation of phantom pain to cortical reorganization. Flor et al.[27] investigated the relation between the degree of cortical reorganization and the severity of the phantom pain in 13 amputees using magnetoencephalography. The amount of phantom limb pain was closely correlated ($r = 0.93$) with the amount of cortical reorganization. Birbaumer et al.[28] studied the effect of local anesthetic block of the brachial plexus on the amount of cortical reorganization. Using somatosensory evoked potentials they demonstrated, first, that the linear shift associated with cerebral reorganization was correlated with the amount of phantom pain, and secondly, relief of pain by the

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anesthetic block was accompanied by a substantial mean reduction or reversal of the amount of linear shift of reorganization. The rapidity with which the plexus block abolished cortical reorganization, reflected a surprising impermanence or plasticity of an apparently long-standing system of reorganization. Davis et al.[29] concluded from microelectrode recordings in the thalamus of amputees, that reorganization in the sensory nucleus offers another mechanism by which stump afferents might influence phantom sensation including pain.

Remarks on the Present Case. The special features in the present case of phantom penile erection include: 1) The penile amputation was complete unlike reported cases in which a stump remained, through which micturition occurred; 2) the presence of a phantom only in the erect state and not in the flaccid state; 3) the replica in the phantom of the preoperative ulcerated lesion with its associated pain; 4) the unusual facility with which an erotic stimulus precipitated a response; 5) a rare instance of bihemispheric cortical representation is exemplified; 6) the 20 year duration of the phenomenon.

The physiology of male tumescence is only partly understood. It can be considered an involuntary, reflex or automatic response to a thought or sensory perception of an erotic nature. The efferent system is autonomic by way of the lower spinal cord parasympathetic and sympathetic outflow. Sensory afferents are carried in the somatosensory system. The erectile mechanism probably involves relaxation of afferent small penile arteries with compression or constriction of venous outflow.[30] In our patient there were probably remnants of the most posterior part of the corpora cavernosa. Presumably under normal circumstances, in humans, erotic stimuli involve the parietal sensory cortex whence the penile vasomotor response is elicited, possibly via a descending hypothalamic pathway. The resultant genito-pelvic sexual feeling could participate in a feedback process through the parietal cortex.

In the sensory homunculus of Penfield and Rasmussen[9] the genitalia are depicted on the medial surface of the hemisphere, in the most inferior position, just below or posterior to the region of the toes. In their electrical exploration of the human cortex, sensory responses referred to the genital region were rare. In one patient, stimulation at two points in the posterior portion of the post-central area near the central fissure, produced a sensation in the contralateral side of the penis. Stimulation of the cortex never produced erotic sensations of any sort. Also, of some relevance here, stimulation never elicited a vasomotor response. An abnormal erotic feeling of cerebral origin is rarely produced by focal epilepsy;[31] it is better known in the Klüver-Bucy syndrome[32] in which the temporal lobes are damaged bilaterally.

Compared with the human upper limb with its remarkable sensorimotor dexterity, the penis is largely a tactile organ whose only response is passive or automatic. In the non-erect state, sensory awareness of the penis is almost nil, a circumstance which, it might be inferred from experience with phantom limbs, would not favor the ready development of a phantom after amputation. Also judging from cortical stimulation studies, the cortical area representing the genitalia is probably small with little scope for cortical reorganization. The concept that the propensity to develop a phantom is related to the size of the area of cortical representation, finds little support here.

The Phantom. Based on the synopsis of up-to-date information concerning phantom limb phenomena, already presented, it may be posited that our patient's phantom erection was also a product of the "deafferented" parietal sensory cortex. There was no stump to complicate the interpretation. The presence of a phantom only in the erect state and not in the flaccid state represents a virtually unique event. Creating a realistic concept of the neural circuitry involved, is an impossibility at present. In attempting a verbal interpretation of the phenomenon, one might say that a thought or a visual perception first gained an erotic connotation. The most likely site for this is the genital area of the parietal sensory cortex. In the absence of the male organ, a cortical sensory image of the normal erect state was evoked. The usual duration of this phase was not sought from the patient. Whether descending autonomic impulses played a part by producing changes in any residual erectile tissue is a matter for speculation. In Heusner's case 1[6] a traumatic paraparesis apparently ended the patient's phantom experience.

For a phantom to make its appearance only under certain physiological circumstances is almost unknown in phantoms of other parts. Phantom limbs may be influenced by mental concentration, emotional states, surprise, pain, wearing a prosthesis, etc., but show no change at all comparable to that of the phantom erection. Breast phantoms may clearly swell premenstrually and slowly recede again after cessation of the period.[33] Along somewhat the same line, Jankovic and Glass[34] described tardive dyskinesia in a phantom arm, as a result of metoclopramide therapy. The occurrence of a phantom only of the erect state may reflect a relatively greater sensory experience in that state, resulting in a more abundant neural connectivity in the parietal cortex.

Whether the sensory cortex is normally subject to analogous changes under the influence of ideas and perceptions cannot be answered. Normally penile erection, a stereotyped reaction, would be sensed in the parietal cortex, where a neural tracery is left. Presumably, when the peripheral sensory receptors are eliminated as in amputation, the particular cortical circuitry lies unused, waiting to participate when the appropriate stimulus arrives. There is the suggestion here, that we are witnessing evidence for the cortical basis of a thought, a simple one, linked to a basic biological function. The thought is tactile, it is vivid, it approaches the form of a tactile hallucination (the phantom). Where else in the human nervous system would an accustomed thought or perception gain an erotic implication, if not in the sensory cortex? This is tantamount to suggesting that, in humans, innate sexual drive and appetite are centered in the parietal cortex where other influences, for example, endocrine can play their part.

Although much of this speculation seems to imply limited neuronal activity locally in the sensory cortex, it must always be regarded as an evanescent focus within a widespread, dynamic, ever-changing, cerebral electrical activity whose "sweep" encompasses every new cerebral interval change, thereby constituting the mind and self-awareness. Regrettably our patient was not asked about the occurrence of ejaculation, whether his erotic inclination could have represented heightened activity and whether any particular pattern was associated with nocturnal dreaming. The effect on the phantom of castration and estrogen therapy used in the treatment of prostatic carcinoma, could have been informative.

The presence in a phantom limb of a pre-existing pain, that is pain that predated amputation, is not uncommon and poses a

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major therapeutic challenge.[35] In the present case, there is the unusual situation that the ulcerated lesion and the associated pain, were transient, reappearing only as part of the erect phantom, not otherwise. The neural tracery corresponding to the ulcer and pain, entered awareness only as part of the phantom. The recurrence of the pain was not just the ordinary recall of a pain; it was an actual re-creation of the original neural tracery (engram). The intimate relationship of the pain to the somatosensory (tactile) phantom, warrants scrutiny for any lessons it might bear concerning the biology of pain. The literature contains no record of a cortical pain homunculus. The same is true of the cortical localization of hotness, without any tactile component. Penfield and Rasmussen[9] reported no pain responses on cortical stimulation and assigned pain to the thalamus. It would seem that pain, being inconstant, does not establish its own homunculus, and gains its localization signature through tactile association. Phantom limbs tend to persist when pain is present. Clinically most pains have a tactile admixture.

Phantom Genitalia in Paraplegia. Weinstein[13] reported that all 150 cases of severe spinal cord injury that he studied, had a phantom of one paralyzed part or another at some time after the injury. In Bors' study[12] of 50 patients with cord injury, 13 reported only non-erect penile phantoms, 8 erect phantoms only and 6 both erect and non-erect phantoms. Of the non-erect group, 9 specified the tip as represented and 3 the shaft, one was undecided. In the erect group the phantom occurred with and without erotic stimulation. In addition, reflex, unfelt actual erections occurred in 43 of the 50 cases. In three cases, erect phantoms ceased after anterolateral chordotomy, an effect which would argue against a wholly cerebral mechanism in paraplegic cases.

Events during dreaming inform us as much about dreams as they do about phantoms, for most paraplegics walk normally in their dreams[12] and male paraplegics may experience penile erections with or without a sensation of ejaculation.[36]

Phantom penis is a very special syndrome even within the field of phantom phenomenology. With a more complete acquisition of historical data than pertained in the present case, there is the potential to gain further insight into the normal and abnormal physiology of high level behavior and pain.

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