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Uterine phantom pain syndrome

Abstract

Phantom uterus pain is an uncommon phenomenon observed in women following a hysterectomy, where painful sensations resembling menstrual cramps or uterine contractions persist despite the removal of the uterus. This report presents the case of a 34-year-old woman from Bogotá, Colombia, diagnosed with advanced endometrial adenocarcinoma who underwent a radical hysterectomy. The patient developed progressive pelvic pain associated with dyspareunia, suggestive of phantom uterus syndrome. Management included neuromodulators and pelvic nerve blockade. This case highlights the complexity of phantom uterus pain, where neuronal plasticity and neuroinflammatory responses may play crucial roles in the generation and persistence of post-surgical pain symptoms. It underscores the need for individualized therapeutic approaches and a deeper understanding of underlying mechanisms to enhance the quality of life of affected patients.

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Keywords: phantom limb pain, endometrial cancer, hysterectomy, phantom uterus, complete pelvic blockade

Introduction

Phantom limb pain (PLP) is defined as pain or discomfort in a part of the body that has been amputated or removed. Although it is classified as neuropathic pain, the pathophysiology is far more complex. It typically occurs in amputated limbs; however, this phenomenon has also been described for amputations involving the tongue, breast, teeth, testicles, penis, bladder, and anus. A literature search in major academic databases (PubMed, EMBASE, OVID, and Google Scholar) revealed only one publication from

1971 referring to phantom uterus [1]. Regarding internal pelvic organs, a case report was found on phantom sensation following correction of uterine prolapse, which was managed with the use of a pessary [2]. Phantom bladder pain following cystectomy was also described, managed with lumbar sympathetic ganglion blockade [3]. Additionally, phantom rectum description was studied in a group of 22 patients with rectal amputation due to neoplastic disease, where 15 patients reported phantom rectum sensation, including gas passage and defecatory urge, with four experiencing pain [4].

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Case report

This report presents the case of a 35-year-old woman from Bogotá, Colombia, diagnosed with adenocarcinoma of the lower uterine segment, moderately differentiated, FIGO 2, nuclear grade 2, with focal squamous differentiation. The tumor infiltrated 90% of the uterine wall and invaded more than 50% of its thickness. Circumferential involvement of the isthmus and submucosal cervical infiltration were identified, with extension to the anterior and posterior uterine walls, reaching the middle third of the uterine body. Immunohistochemical studies showed positive reactivity for cytokeratin (CK), paired box (PAX), estrogen receptors (ER), progesterone receptors (PR), P16, vimentin, and high-weight CK in areas with squamous differentiation. Beta-catenin was found to be mutated with nuclear and cytoplasmic expression, and the Ki67 proliferation index was 50%. The tumor was negative for protein 53 (P53) and carcinoembryonic antigen (CEA). Molecular testing for microsatellite instability (MSI) revealed intact nuclear reactivity in postmeiotic segregation increased 2 (PMS2), MLH1, MSH2, and MSH6. The tumor measured 7 × 4 cm on the surface and exhibited lymphovascular space invasion (LVSI). However, the parametrial margins and vaginal cuff were free of tumors. The fallopian tubes and ovaries showed no tumor involvement, with normal findings of white bodies and follicular cysts. Her surgical history includes closure of a congenital atrial septal defect (ASD) at age 13, conization and laparoscopy for a right ovarian cyst in 2014, and an appendectomy in 2017.

Her gynecological history includes menarche at age 13, regular menstrual cycles, initiation of sexual activity at age 18, no use of contraceptive medications, and nulliparous with no history of pregnancies. She underwent surgical management with a radical hysterectomy, bilateral salpingo-oophorectomy, and pelvic and paraaortic lymphadenectomy via laparotomy in 2022, followed by adjuvant radiotherapy IMRT. A total dose of 45 Gy was delivered in 23 fractions (1.8 Gy per session) to the pelvis (PTV45). Additionally, a boost was administered via high-dose-rate (HDR) intracavitary brachytherapy after completing 36 Gy of external radiotherapy. During and after treatment, the patient experienced expected side effects, including nausea, emesis, and pelvic pain, along with cystitis, which was successfully managed with phenazopyridine.

The patient was referred to the palliative care service in June 2024. During the evaluation, she reported experiencing cramp-like pain in the pelvic region for a year and a half. Initially weekly, the pain had become

daily over the last 6 months, occurring 3 times a day, with crises lasting approximately 5 to 10 minutes. The pain was similar to menstrual pain experienced before the hysterectomy. In addition to this pain, the patient reported symptoms such as increased appetite and breast pain as if she were menstruating. She managed the pain with self-prescribed hyoscine and ibuprofen or plain hyoscine, achieving relief, with a maximum pain score of 6/10 on the numeric rating scale (NRS). After medication, the pain score decreased to 2/10 with analgesic effects lasting up to 12 hours.

Emotionally, the patient, who intended to have children, expressed adequate coping with the change in her life plans. She also reported relationship issues due to dyspareunia associated with the described symptoms. Physical examination revealed a median infra umbilical abdominal scar with keloid, tenderness on palpation in the left iliac fossa, without signs of peritoneal irritation, and pain on vaginal examination. A pelvic computed tomography (CT) scan was performed, showing the surgical absence of the uterus and adnexa without any evidence of tumor recurrence or other causes explaining the symptoms. Considering the patient's clinical history and the neuropathic and visceral characteristics of the pain, despite its rarity, the possibility of phantom uterine pain syndrome was considered. The patient was treated with the neuromodulator duloxetine, as she did not tolerate pregabalin due to drowsiness, and she underwent sympathetic blockade of the superior hypogastric plexus with favorable outcomes post-procedure, with an NRS of 0/10 one week after the blockade.

Discussion

Phantom limb pain (PLP) occurs after the amputation of a limb or organ, with limbs being more commonly affected than visceral organs. The explanation of the etiology of phantom limb sensations and their pain is divided into peripheral mechanisms, including axonal damage, inflammatory changes, the presence of neuromas, and peripheral sensitization; and central mechanisms, including central sensitization processes, the neuromatrix theory, and cortical reorganization [5].

Although visceral pain is diffuse and difficult to describe, some organs like the uterus have a specific sensory experience associated with menstruation, pregnancy, and childbirth, leading to a sense of the pelvic organ and its memory. Phantom uterus pain has been described as sensations of cramp-like pain following a hysterectomy, which is equal in duration, intensity, frequency, and characteristics to menstrual

cramps and labor pain associated with uterine contractions. Dopart prefers to specify that this phenomenon is more accurately described as phantom uterine contraction pain rather than the sensation of a phantom uterus, as it is a different experience from that of an amputated limb [1]. Phantom pain in external organs might be related to a kinesthetic experience related to the structure, whereas in internal visceral organs, it is related to function. Thus, phantom pains in the bladder, rectum, and uterus have been associated with dysuria and tenesmus; defecation, flatulence, and secretion; and colic and contractions, respectively [1].

The uterus is innervated by the uterovaginal nerve plexus, which originates from the inferior hypogastric plexus, derived from the superior hypogastric plexus. This plexus contains sympathetic and parasympathetic efferents, and visceral afferents [6]. Visceral pain includes autonomic sensations, so sympathetic activation in visceral pain, with increased adrenergic activity, contributes to its chronicity. Therefore, in relation to PLP, the autonomic nervous system plays a key role in symptoms of dysautonomia and phenomena of hyperalgesia and allodynia. This is explained by increased peripheral nerve response to catecholamines, greater expression of alpha-1 adrenergic receptors, and sympathetic bursts in the dorsal root ganglion [7]. All of this explains the clinical usefulness of superior hypogastric plexus blockade as performed in the presented case.

It is essential to establish a differential diagnosis between radiotherapy side effects and pain associated with phantom uterus syndrome. In this case, symptoms such as nausea, emesis, and pelvic pain were consistent with expected radiotherapy side effects. Cystitis was confirmed and effectively treated, indicating a radiotherapy-related origin. Persistent pelvic pain was evaluated through pelvic CT imaging, which ruled out tumor recurrence or other structural causes.

The treatment of PLP is based on pharmacological strategies, with neuromodulators being the cornerstone as this is considered neuropathic pain. Gabapentinoids, tricyclic antidepressants, dual serotonin-norepinephrine reuptake inhibitors, opioids, and ketamine have been described for use. Non-pharmacological strategies include non-invasive techniques [virtual reality, graded motor imagery, transcutaneous electrical nerve stimulation (TENS), transcranial magnetic stimulation, and direct current stimulation], psychological approaches (cognitive-behavioral therapy and hypnosis), and interventional pain management (electrostimulation in the dorsal

root ganglion and peripheral nerves, diagnostic and neurolytic sympathetic blockades, and peripheral nerve blockades) [5].

In conclusion, phantom uterus pain is a rarely described entity in the literature. Understanding PLP in visceral organs involves comprehending the autonomic nervous system and its role in chronic neuropathic pain. Identifying it requires investigating visceral symptoms that allow for a sensory awareness of an internal organ, which is reported when visceral phantom pain occurs. Specific treatment has been little explored but follows general PLP strategies, including pharmacological approaches focused on neuromodulators, and non-pharmacological methods involving non-invasive and invasive techniques, with particular emphasis on sympathetic and peripheral nerve blockades. Non-pharmacological techniques from physiotherapy that involve pelvic floor rehabilitation are also important, alongside evaluating and treating psychological and emotional factors that may contribute to pain chronicity.

Conclusions

Phantom uterine pain is an underrecognized entity that reflects the complexity of neuropathic pain in visceral organs, where the interaction between the central and autonomic nervous systems plays a crucial role. Understanding this phenomenon is essential for developing comprehensive therapeutic strategies that address both the physical and emotional aspects of pain. It is imperative to encourage research that explores the underlying mechanisms and the most effective interventions, allowing for personalized treatments and improving the quality of life for patients affected by this syndrome.

Article information and declarations

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Ethics statement

This case report has been approved by the Ethics Committee of the National Cancer Institute of Colombia, ensuring compliance with the corresponding ethical principles and regulations. Written informed consent was obtained from the patient for the publication of the information in this report, guaranteeing her privacy and confidentiality.

Author contributions

Participated in the idea, writing, and review of the manuscript, as well as in the conceptualization and design of the case report — FAL-A; contributed to the conceptualization and design of the case report and the writing of the manuscript — SMB-P; participated in the writing of the manuscript — NB-J, FAM-M, and EM-N. All authors reviewed and approved the final version of the manuscript for publication.

Conflict of interest

The authors declare that there are no financial, personal, academic, or any other conflicts of interest in relation to this article. No external funding or institutional support was received that could influence the results and conclusions presented in this work.

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Supplementary material

None.

References

1. Dorpat TL. Phantom sensation of internal organs. *Compr Psychiatry*. 1971; 12(1): 27–35, doi: [10.1016/0010-440x\(71\)90053-8](https://doi.org/10.1016/0010-440x(71)90053-8), indexed in Pubmed: [5540721](https://pubmed.ncbi.nlm.nih.gov/5540721/).
2. Solano Sánchez RG, García Alonso F, Maldonado Miranda M. Sensación fantasma posterior a la reparación quirúrgica de un prolapso de órganos pélvicos estadio IV. *Anales Médicos*. 2017; 62(4): 271–274.
3. Park KE, Cheon KS, Ok SH, et al. Phantom bladder pain. *Korean J Anesthesiol*. 2012; 63(4): 376–377, doi: [10.4097/kjae.2012.63.4.376](https://doi.org/10.4097/kjae.2012.63.4.376), indexed in Pubmed: [23115696](https://pubmed.ncbi.nlm.nih.gov/23115696/).
4. Ovesen P, Krøner K, Ørnsholt J, et al. Phantom-related phenomena after rectal amputation: prevalence and clinical characteristics. *Pain*. 1991; 44(3): 289–291, doi: [10.1016/0304-3959\(91\)90099-J](https://doi.org/10.1016/0304-3959(91)90099-J), indexed in Pubmed: [2052399](https://pubmed.ncbi.nlm.nih.gov/2052399/).
5. Boomgaardt J, Dastan K, Chan T, et al. An algorithm approach to phantom limb pain. *J Pain Res*. 2022; 15: 3349–3367, doi: [10.2147/JPR.S355278](https://doi.org/10.2147/JPR.S355278), indexed in Pubmed: [36320223](https://pubmed.ncbi.nlm.nih.gov/36320223/).
6. Huntzinger J, Selassie M. Interventional pain management in the treatment of chronic pelvic pain. *Curr Urol Rep*. 2023; 24(4): 165–171, doi: [10.1007/s11934-022-01141-6](https://doi.org/10.1007/s11934-022-01141-6), indexed in Pubmed: [36719535](https://pubmed.ncbi.nlm.nih.gov/36719535/).
7. Arslan D, Ünal Çevik I. Interactions between the painful disorders and the autonomic nervous system. *Agri*. 2022; 34(3): 155–165, doi: [10.14744/agri.2021.43078](https://doi.org/10.14744/agri.2021.43078), indexed in Pubmed: [35792695](https://pubmed.ncbi.nlm.nih.gov/35792695/).