

Phantom Tongue Pain: A Case Report

Grant Drake, DO, Eric Lam, DO, Benjamin Cooper, MMS

Abstract

Case Diagnosis

We present a case on phantom tongue pain observed in a 65-year-old male with a history of adenoid cystic carcinoma with involvement of the base of the tongue and supraglottic laryngopharyngeal cancer status-post laryngopharyngectomy and glossectomy.

Case Description

A 65-year-old Hispanic male was admitted to our acute inpatient rehabilitation facility for functional decline after undergoing a total laryngopharyngectomy and glossectomy. Several days into his stay the patient began to describe a pain sensation that his tongue was still present in his oropharynx. He began participating in mental imagery exercises with physical therapy consisting of mentally visualizing moving the tongue through the full range of motion. This was performed during therapy sessions and independently by the patient while in bed. At the end of his stay, the patient reported a decrease in pain.

Discussions

Phantom limb pain is a poorly understood pain syndrome which involves a patient experiencing pain appearing to come from a limb which is no longer present. Proposed mechanisms involve neurogenic change centrally and near the site of amputation including Wallerian degeneration and collateral sprouting. Current treatment options include anticonvulsant medications and therapeutic desensitization techniques including mirror therapy and mental imagery techniques. Post total glossectomy phantom pain is rare, and as this is a singular appendage, current techniques that rely on the presence of an intact limb, such as mirror therapy could not be applied to our patient. Therefore mental imagery techniques originally developed for extremity amputation required adaptation to the context of total glossectomy.

Conclusions

In our case, movement imagery training was adapted to a singular appendage amputation and resulted in a decrease in the reported pain score. Novel approaches to alleviating phantom pain, like virtual reality, should be reevaluated for effectiveness in relieving pain. Utilizing therapeutic desensitization techniques may allow for direction of neuroplasticity in order to decrease pain.

Introduction

- The incidence of phantom limb pain (PLP) varies in literature, but may be as high as 95 percent.^{1,2} Phantom pain is typically described as burning, aching, or electrical shooting pain in the amputated limb.¹ Its mechanism of action is poorly understood. There are several accepted pharmacological therapies that can be effective in relieving phantom pain.
- Phantom tongue pain can be treated using a variety of pharmacological interventions; however, the clinically relevant outcomes in studies, like pain, mood, function, depression, and quality of life vary based on the treatment provided.³ Morphine continues to be an effective drug of choice for short-term analgesia. Ketamine and Gabapentin appear to be beneficial.³ Mirror therapy is one novel type of treatment that has been used with success, in treating phantom limb pain.⁴ Although phantom pain might be eliminated with these therapies, the phantom sensation may persist despite the use of pharmacological interventions.
- Phantom tongue pain is a condition that is rarely reported, with there being only one known case in literature, published back in 1976.⁵ Our case uses currently accepted practices for the treatment and management of phantom limb pain and applied it to our rare case of phantom tongue pain.

Case Description

A 65-year-old Hispanic male was admitted to our acute inpatient rehabilitation facility for functional decline. The patient had been diagnosed with adenoid cystic carcinoma involving the base of tongue and supra-glottic larynx several months prior. He underwent total laryngopharyngectomy, glossectomy and neck dissection with reconstructive thigh flap placement approximately 1 week prior to admission to our facility. A PEG tube had been placed, and he was to remain NPO until his surgical follow up.

On admission the patient was aphonic, but able to communicate by dry erase board. His primary complaint was of severe neck pain, he denied fever, chill or other associated symptoms. Initial physical examination of the neck showed circumferential swelling and erythema. No evidence of purulence or drainage was visualized, surgical site appeared well healing with no dehiscence. He experienced pain on neck rotation but was able to demonstrate full range of motion.

He was receiving duragesic patch 50 mcg q72h, Percocet 5-325 mg q8h prn for breakthrough pain and gabapentin 300mg q8h. He was seen by the pain management consultation service and his analgesic regimen was adjusted, duragesic patch was discontinued and he was started on oxycontin 10 mg q12 with percocet 5-325mg 1-2 tablets q4h for breakthrough pain.

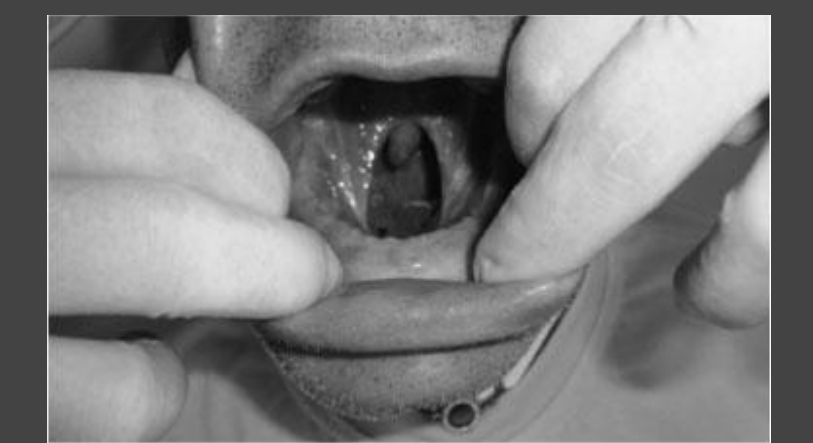
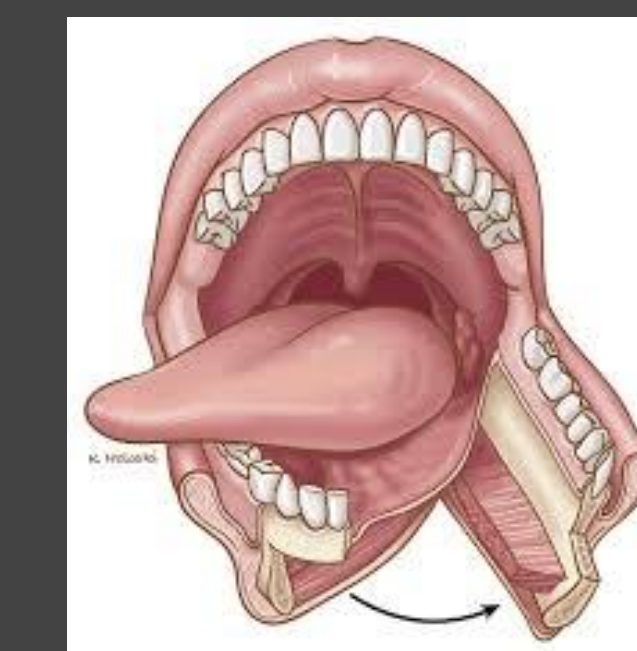
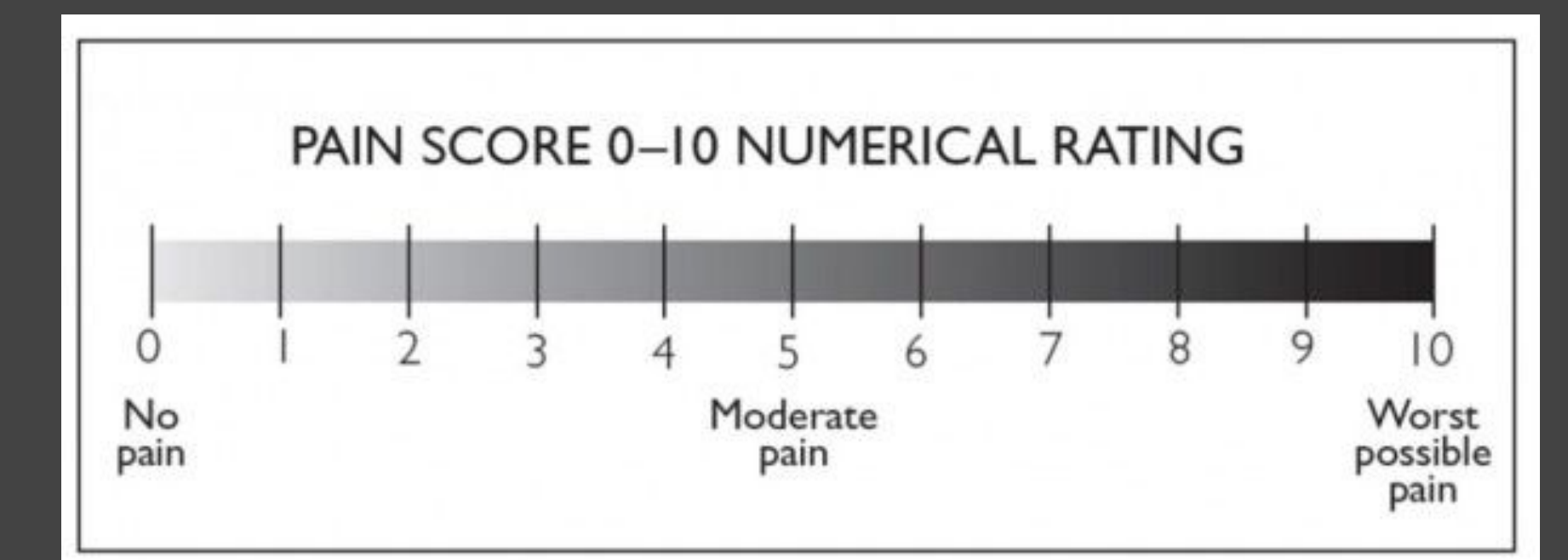
Several days later, his generalized neck pain was well controlled, however on interview the patient described a sensation that his tongue was still present in his oropharynx and began to complain of tongue pain, initially at the 7-8/10 level per NRS-11 numeric pain scale. Physical examination revealed only expected surgical changes in the oropharynx, laboratory investigations were unremarkable.

The patient's gabapentin dosage was increased to 400 mg TID and he began participating in mental imagery exercises with physical therapy consisting of mentally visualizing moving the tongue through a range of motion consisting of elevation, depression and lateral movements, he would then visualize isometric holds of the tongue in each of those positions. He was to perform this for 10 repetitions with therapy and later while in bed up to once per hour.

The patient performed these mental imagery exercises with physical therapy and independently while in bed over the course of one week of his inpatient stay. At the end of his stay, the patient reported a decrease in pain to the level of 5/10 per NRS-11 numeric pain scale.

Discussions

- Due to the complex nature of pain generation in the phantom limb pain patient, a multi-disciplinary approach to treatment is now preferred, including pharmacologic intervention and therapeutic modalities. Current therapeutic techniques for phantom limb pain include: mirror therapy, desensitization techniques, graded motor imagery or movement imagery training. The pharmacologic approach relies on medication classes traditionally utilized in the treatment of neuropathic pain such as anticonvulsants or tricyclic antidepressants.
- As a physiatrist, add ressing the complexity of phantom limb pain is an essential component of the amputation rehabilitation process. The more common amputations such as transmetatarsal, transtibial, transfemoral, or amputations involving the upper extremities have proven treatment approaches; whereas, treating singular appendage amputations, such as the tongue, are still being researched. Within this nascent field of study, the number of patients with this presentation is quite small, which makes evaluating treatment modalities limited.
- Phantom limb pain is a poorly understood pain syndrome with proposed central as well as peripheral neuropathic mechanisms. Investigational studies such as *Motor reorganization after upper limb amputation in man. A study with focal magnetic stimulation* by Cohen et al. demonstrated neuroplastic changes in patient's status post limb amputation.⁶ Nerves reaching the site of amputation undergo axonotmesis, which may then lead to wallerian degeneration. Additionally, collateral sprouting from fibers near the amputation site may also lead to hyperalgesia and other sensory changes including "expansion of peripheral receptive fields".
- Mirror therapy was first used in 2004 as a treatment for PLP in people with a lower limb amputation.⁴ It was used with success and has been considered an effective adjunct to medical therapy for patients undergoing an amputation of their limb.⁸ In one study, patients using mirror therapy practiced for 4 weeks showed a decrease in PLP pain severity.⁸ This study further showed that patients who were not using prosthesis had a higher benefit from mirror therapy than those with prosthesis.⁸ Since our patient was without a tongue, mirror therapy was unable to be used. Therefore, our patient's treatment plan included desensitization and mental imagery, and its use contributed to the reduction of pain seen in our patient's phantom tongue pain.
- Mirror therapy consists of a mirror being placed adjacent to the intact limb and then moved in exercises designed to influence reorganization of the cortex with this visual input. The concept of being able to influence the remodeling of the cortex with visual inputs allows for the basis that afferent stimuli to the cortex can bring about neurological change. Mental imagery, if vivid enough and regularly repeated, could possibly be just as effective as mirror therapy. An area for further research would be virtual reality models for bilateral limb amputations or tongue amputations that could provide visualization of the missing appendage, which is not possible with mirror therapy. It would be important to distinguish if mental imagery is as efficacious, if not more so, compared to visual inputs and determining if there are any shortcomings to mental imagery in the treatment of phantom limb pain.



Conclusions

- Current therapeutic approaches such as mirror therapy and movement imagery training are thought to direct the neuroplastic potential of the amputee toward remodeling of afferent and efferent pathways in favor of somatosensory "acceptance" of the altered anatomy. These approaches are typically performed in the context of a limb amputation. In our case, movement imagery training was adapted to a singular appendage amputation and resulted in a decrease in the patient's reported pain score. Novel approaches to alleviating phantom pain, like virtual reality, should be reevaluated for effectiveness in relieving pain. Moreover, there has been no general consensus on the optimal length of mirror therapy treatment.
- There are a wide variety of modalities for treating phantom limb pain that involve both pharmacological and nonpharmacological treatments. Taking advantage of the plasticity of the central and peripheral nervous system allows for therapies such as mirror therapy and repetitive mental imagery to promote a rewiring of the pain pathways involved in phantom limb pain.

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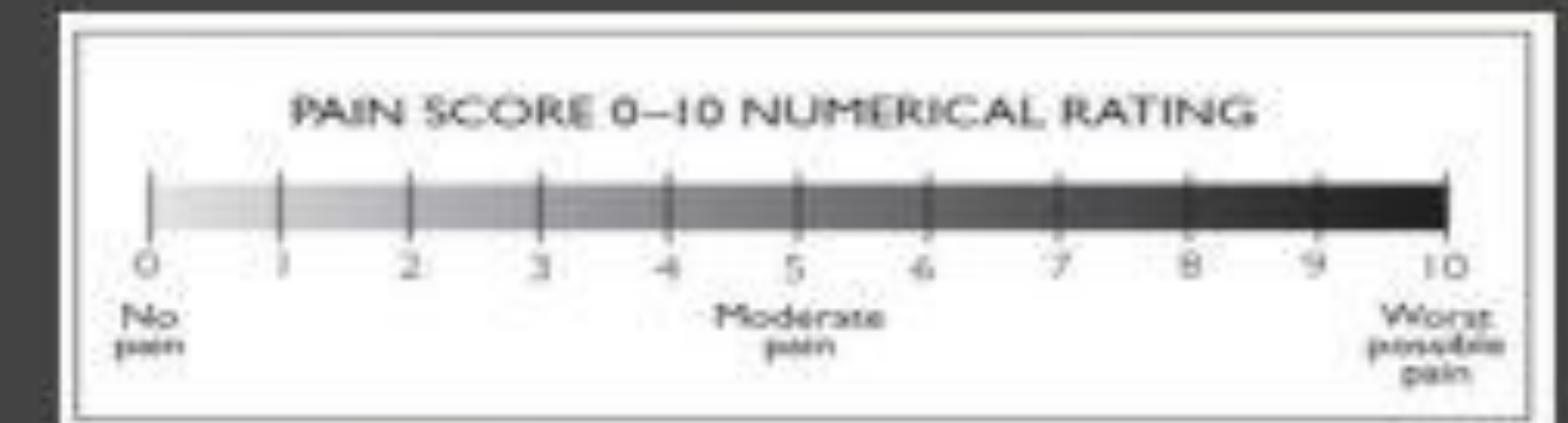
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