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CASE REPORT

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Unilateral tongue atrophy as the initial clinical manifestation in a patient with prostate cancer

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Key Clinical Message

Unilateral tongue atrophy can be a rare and crucial early indicator of metastatic prostate cancer, highlighting the need for vigilant monitoring in clinical assessments. This case underscores the importance of considering cranial nerve involvement, especially the twelfth, for timely intervention and comprehensive patient care.

Abstract

Prostate cancer, ranking among the most prevalent cancers, often manifests with skeletal metastases. Cranial nerve involvement, particularly the twelfth cranial nerve (XII), as an initial presentation is exceptionally rare. This case report outlines a unique instance of unilateral tongue atrophy as the primary clinical manifestation in a patient diagnosed with metastatic prostate cancer. A 54-year-old man presented with dysarthria and progressive weakness, later revealing signs of hypoglossal nerve paralysis, unilateral tongue atrophy, and skeletal metastases involving the base of the skull. Imaging studies, including CT and MRI, confirmed diffuse lytic lesions and cranial nerve entrapment. Further investigations identified elevated PSA levels, confirming acinar prostate adenocarcinoma. The patient underwent hormone therapy due to the poor prognosis. Prostate cancer's skeletal metastases are well-documented, but cranial nerve involvement remains rare, particularly with isolated XII nerve manifestation. The discussion emphasizes the diagnostic challenges, imaging techniques' roles, and the impact on prognosis and quality of life. This case underscores the rarity of unilateral XII nerve involvement as the initial presentation of metastatic prostate cancer. Clinicians should consider this manifestation, especially in men over 40, warranting a thorough diagnostic approach, including PSA measurement and referral for appropriate oncological and urological interventions.

KEYWORDS

acinar prostate adenocarcinoma, cranial nerve involvement, hypoglossal nerve paralysis, prostate cancer, skeletal metastases, unilateral tongue atrophy

1 | INTRODUCTION

Oncological diseases continue to pose a significant burden on public health. Prostate cancer ranks as the third most common cancer overall and the second when differentiated by gender.[1] This cancer often exhibits a high metastatic capacity, particularly to the skeletal system, with metastatic rates reaching up to 82%, accompanied by elevated PSA levels compared to other metastases.[2] Although the spine, pelvis, and femur are common sites, involvement of the skull base, leading to cranial neuropathies, is rare.[3] Unusually, the involvement of cranial nerves, particularly the twelfth cranial nerve (XII), has been described as an uncommon and rare initial manifestation of prostate cancer,[4] which is the focus of our case.

2 | CASE PRESENTATION

A 54-year-old man, with no oncological family history and no known pathological personal history, presented to the neurology service with dysarthria and progressive weakness over the last 5 months. Dysarthria affected speech and tongue biting on the left side, with reduced mobility. Progressive weakness started proximally, then distally, in all four limbs over 2 months. Additionally, he experienced lumbar vertebral pain, which extended throughout the spine within a month. Subsequently, urinary symptoms developed (urinary urgency, nocturia, and altered final urinary dribble) and significant weight loss (27 kg in 5 months).

During the physical examination of the patient, a general appearance indicative of illness was noted, accompanied by mild mucocutaneous pallor and signs of Grade I malnutrition. Dysarthria was evident. isolated paralysis of the hypoglossal nerve, unilateral atrophy of the left half of the tongue, and deviation to the left on protrusion and retraction to the right on intrusion (Figure 1A). Additionally, weakness in all four extremities was observed, predominantly affecting proximal regions. Muscular weakness was assessed using the Daniels strength scale, revealing a score of 3/5, with no evidence of other notable abnormalities. Imaging studies, including computed tomography (CT), revealed diffuse lytic lesions at the skull base and bilateral involvement of the hypoglossal canal, predominantly on the left side (Figure 3B). Brain MRI demonstrated increased fat in the left half of the tongue with hyperintensity and enhancement suggestive of atrophy due to denervation (Figure 1B), raising initial suspicions of metastatic lesions. Ultrasound identified Grade IV prostatic growth, with a PSA level measuring 4828 ng/mL. Biopsy confirmed acinar prostate adenocarcinoma, a Gleason score of 7 in the right lobe and a Gleason score of 9 in the left lobe, with OMS/ISUP grading of 3 for the right and 5 for the left (Figure 2). Bone scintigraphy revealed a hyperscan pattern with 90% tracer retention (Figure 3A). Due to the poor prognosis and therapy availability, our patient underwent chemotherapy without undergoing a routine adenoidectomy.

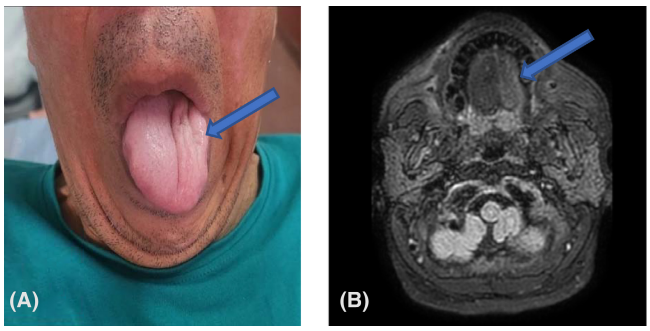

FIGURE 1 (A) Patient with left tongue atrophy, arrow indicating protrusion (Image obtained with the patient's consent in the Neurology Department of Manolo Morales Peralta Hospital, Managua, Nicaragua). (B) Magnetic resonance imaging (MRI) of the brain in axial cut, Flair sequence. (B) Hyperintensity is observed in the left half of the tongue, highlighting marked atrophy (indicated by the arrow). This study was conducted using a 3T Philips MRI machine and was performed on August 29, 2023 at the Manolo Morales Peralta Hospital.

3 | METHODS

Within our clinical methodology, we systematically substantiated the case, initiating with the clinical approach to define syndromes. These encompassed unilateral left-sided cranial nerve XII mononeuropathy, four-limb weakness syndrome, irritative and obstructive urinary symptoms, constitutional manifestations, and vertebral pain. Considering the temporal evolution, patient demographics, and age, we prioritized a neoplastic etiology as the primary consideration. Following this preliminary assessment, we conducted paraclinical investigations, involving comprehensive general laboratory analyses and imaging modalities, culminating in the definitive identification of metastatic prostatic adenocarcinoma.

We undertook an exhaustive review of diverse differential diagnoses, methodically excluding them based on clinical presentation and imaging studies. Various syndromes associated with cranial nerve involvement, such as Collet–Siccard syndrome, Villaret syndrome, occipital condyle syndrome, and jugular foramen syndrome, have been well-documented.[5] The incorporation of imaging modalities, including CT and MRI, affords a holistic diagnostic perspective. CT provides pivotal insights into bone involvement, surpassing MRI in this aspect, while MRI

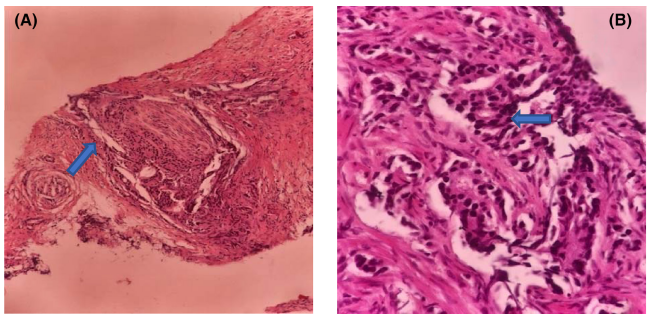

FIGURE 2 Prostatic acinar adenocarcinoma. (A) A 100× magnification, perineural invasion of prostatic acinar adenocarcinoma is evident. (B) 400× magnification, small and poorly formed prostatic glands are observed. The cells exhibit a monotonous appearance, hyperchromatic nuclei, and a high nucleus-to-cytoplasm ratio (images obtained at the pathology Department of Manolo Morales Peralta Hospital, Managua, Nicaragua).

delineates nerve and innervated structure involvement through multiple sequences.[6] Cranial nerve metastasis is an uncommon occurrence within the spectrum of intracranial metastases. To date, only a limited number of cases have been reported concerning secondary cranial neuropathy associated with prostate cancer, with nerves II, III, IV, VI, VII, VIII, and XII being the most frequently affected.[4]

Regarding our patient's therapy, he was assessed by the medical oncology service. Considering the diagnosis of metastatic prostate cancer, a conservative medical approach was chosen, involving chemotherapy with Docetaxel for a total of 12 cycles. Currently, he has completed 4 cycles (one every 2 weeks). Additionally, he received a gonadotropin-releasing hormone agonist (bicalutamide), and an improvement in the Karnofsky performance status scale has been observed.

4 | DISCUSSION

Prostate cancer represents the most common cancer in men, with 90% being acinar adenocarcinomas graded according to the Gleason scale.[7] Central nervous system (CNS) metastases from prostate cancer are rare, accounting for 0.2%–2% of all CNS metastases.[4] Metastases typically spread to lymph nodes and bone tissue,[8] with a predilection for the skeletal system, causing pain and pathological fractures as expected initial manifestations.[5,9] Skull base metastases often present silently until cranial nerve entrapment occurs, resulting in diverse clinical manifestations depending on the affected nerve, most commonly affecting the optic nerve. Multiple syndromes related to cranial nerve involvement have been described, including Collet-Siccard syndrome, Villaret syndrome, occipital condyle syndrome, and jugular foramen syndrome,

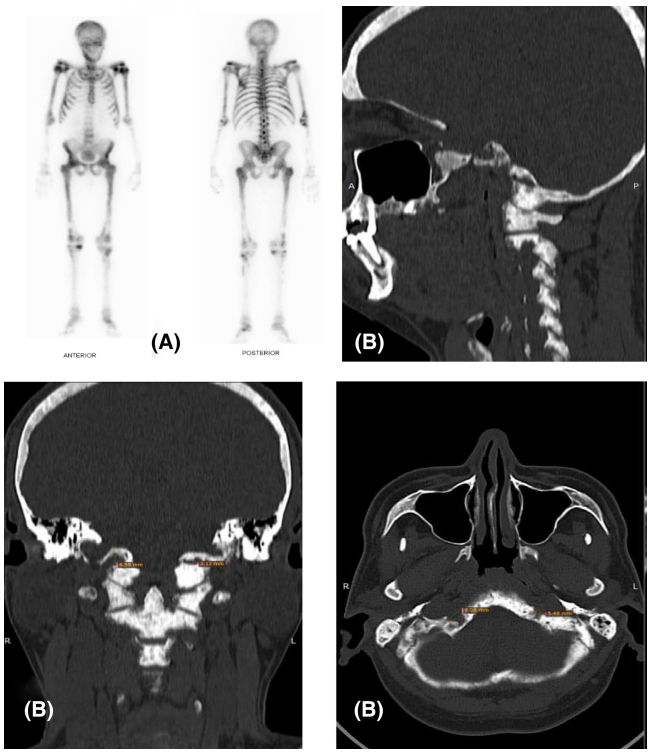

FIGURE 3 (A) Bone scintigraphy in anterior and posterior views. The visualization reveals diffuse uptake in the axial and appendicular skeleton, as well as the cranial vault. These findings are consistent with disseminated secondary involvement, demonstrating a classic pattern of hyperscanning with 90% of the tracer retained in the bone marrow. Importantly, no tracer elimination is observed at the renal level (bone scan, code 043-43, performed on September 7, 2023, at the Nuclear Medicine Department of the National Center for Radiotherapy, Managua, Nicaragua). (B) In the axial, sagittal, and coronal bone window sections of the computed tomography scan, we observed diffuse involvement with lytic and blastic lesions in the bone was observed, including the skull base. Specifically, at the region of the hypoglossal canal, a reduced amplitude was demonstrated on the left side, measuring 3.12 mm, compared to the right side, which showed 6.59 mm in sagittal section. This difference explains the entrapment of the hypoglossal nerve. (A Philips 128-slice CT scanner was used, and the scan was performed on August 28, 2023 at the Radiology Department of Manolo Morales Peralta Hospital, Managua, Nicaragua.)

among others.[5] The combination of imaging techniques, such as CT and MRI, provides a comprehensive diagnostic scenario. CT contributes data on bone involvement, surpassing MRI in this aspect, while MRI depicts nerve and innervated structure involvement through various sequences.[6] Cranial nerve involvement by metastasis falls under intracranial metastases and is considered exceptionally rare. To date, very few cases have been reported on secondary cranial neuropathy due to prostate cancer, with nerves II, III, IV, VI, VII, VIII, and XII being the most commonly affected.[4]

TABLE 1 A table summarizing previous reports in such rare entity.

Year	Author	Title	Associate with prostate cancer	Unilateral	Reference
1996	Keane	Twelfth-nerve palsy. Analysis of 100 cases	No	-	10
1996	Rotta and Romano	Skull base metastases causing acute bilateral hypoglossal nerve palsy	Yes	Bilateral	11
1999	Long and Husband	Features of unusual metastases from prostate cancer	Yes (1/508 patients)	-	3
2002	Capobianco et al.	Occipital condyle syndrome	Yes (4/11 patients)	Unilateral	12
2004	McDermott et al.	*Cranial Nerve Deficits in Patients with Metastatic Prostate Carcinoma*	Yes (2/15 patients)	-	5
2011	Abdullah et al.	Atrophy of the Tongue as the Presenting Feature of Metastatic Prostate Cancer	Yes	Unilateral	6

Our clinical case is based on the exceptional unilateral involvement of the left XII cranial nerve, without additional involvement that could categorize it under multiple cranial neuropathy syndromes or occipital condyle syndrome due to the absence of pain, emphasizing the rarity of this presentation. A review of the literature in Pubmed revealed only a few case reports of unilateral involvement of the XII cranial nerve in patients with prostate cancer and CNS metastasis.

It is crucial to note that despite its rarity, cranial neuropathy significantly impacts the quality of life of prostate cancer patients.[4–6] The development of cranial neuropathy has been considered a sign of poor prognosis in prostate cancer patients. However, its prognosis currently depends on various factors and therapeutic strategies.

Advanced search terms with MeSH keywords, including "Neoplasia," "Prostate Cancer," "XII Cranial Nerve," "Paralysis," and "Nerve Entrapment." The gathered data were summarized in a table, showcasing the year, author, title, association with prostate cancer, lateralization, and the corresponding reference. (Table 1).

According to local epidemiology, this is the first reported case in our country.

5 | CONCLUSION

Cranial neuropathy as the initial manifestation of metastatic prostate cancer is rare, and the isolated presentation of cranial mononeuropathy is even more exceptional. When it presents in men over 40 years, this manifestation should raise suspicions of skull base metastasis, justifying a diagnostic approach that includes PSA measurement and referral to oncology and urology services for appropriate follow-up.

In the latest follow-up evaluation in the neurology outpatient clinic on December 06, 2023, improvement in dysarthria was observed. There was apparent improvement in overall muscle trophism, even at the level of the tongue, with enhanced muscle strength rated at +4/5 according to the Daniels scale, and an improvement in the Karnofsky performance status scale has been observed.

AUTHOR CONTRIBUTIONS

Mohammed Zahran: Investigation; methodology; supervision; writing – original draft; writing – review and editing. **Henry Larios:** Conceptualization; writing – review and editing. **Victor Rosales-Obregón:** Conceptualization; writing – review and editing. **Adrian Coulson Coulson:** Project administration; resources.

FUNDING INFORMATION

We regret to inform that we currently do not have any funding to report for this submission.

CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest to declare.

DATA AVAILABILITY STATEMENT

We commit to ensuring the availability and confidentiality of the information related to the presented clinical case. Our priority is to safeguard patient privacy and adhere to the highest ethical standards in handling medical information. All clinical documentation will be protected and used solely for educational and academic discussion purposes.

CONSENT STATEMENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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