



Case Reports & Case Series

Obstructive sleep apnea secondary to cervical spine chordoma

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ABSTRACT

We present a rare case of obstructive sleep apnea (OSA) secondary to chordoma involving cervical spine with confirmative histopathological diagnosis following the suggestive findings of computed tomography and magnetic resonance imaging. The patient underwent en bloc removal of the tumour involving the vertebral body of C4 with preservation of the nerve roots and vertebral arteries involved, associated with postoperative radiation therapy. Clinicians should be aware that this is a very rare secondary cause of OSA caused by chordoma involving the spine and imaging of the cervical spine may be necessary to avoid incorrect treatment approaches such as CPAP or BiPAP machine in such cases.

1. Introduction

Chordomas of the spinal column are rather rare malignant tumours in the cervical region and they are difficult to treat because of their propensity to local aggressiveness, despite adjuvant therapy [1]. Diagnosis of cervical chordoma is based on radiological examinations, but it is sometimes difficult to establish in contrast to thoracolumbar chordoma and clival chordoma because of its unique feature with an anatomical transition from the thoracic spine to the clivus [1]. To the authors' knowledge, there is no report of cervical spine chordoma secondary to sleep apnea in the pertinent literature. Here the authors report first case of obstructive sleep apnea (OSA) secondary to chordoma involving cervical spine with confirmative histopathological diagnosis following the suggestive findings of computed tomography (CT) scan and magnetic resonance imaging (MRI).

2. Case report

A 46 years old male patient was presented with the complaints of neck pain, headache, pain and numbness in the left arm, together with severe dysphagia, hoarseness, sleep disorder, snore, and fatigue. His past history revealed that he was treated using levothyroxine with a diagnosis of multinodular goitre using ultrasonography (US) and biopsy seven years prior to presentation by an endocrinologist with complaint of throat discomfort. Four years later, complaints of snore, fatigue, and sleep disorder started; then, a diagnosis of OSA was made with polysomnography and he was treated using CPAP (continuous positive airway pressure) auto machine in our hospital at first and then BiPAP

(bilevel positive airway pressure) machine in another center because of the lack of improvement in his complaints. However, his complaint of sleep disorder progressed; difficulty swallowing and hoarseness started together with the complaints of pain and numbness in the left arm, neck pain before admission to our clinic.

A whole-body [¹⁸F]-fluorodeoxyglucose positron emission tomography CT scans showed a hypermetabolic large lesion extending from C2 to C6 in the cervical region with evidence of destruction of C4 vertebral body (Fig. 1a, b). CT scan and MRI of the cervical spine with a contrast medium confirmed the prevertebral heterogenous mass lesion compressing both prevertebral structures and the cervical spinal cord (Fig. 1c–f). An anterior transcervical biopsy procedure was performed by other physician and demonstrated chordoma. Then, the patient subsequently underwent anterior discectomies at C3–4 and C4–5, followed by en bloc removal of the tumour involving the vertebral body of C4 and subtotal removal of the tumour tissue with preservation of the nerve roots and right/left vertebral artery involved. The spine was reconstructed using an expandable corpectomy cage, allograft, and a plating from C3 to C5. Three months later, she received postoperative conventional radiation therapy in another hospital. At 12-month follow-up, he remained disease free without use of CPAP or BiPAP device and neurologically intact and postoperative radiographs demonstrated good position of the construct. A follow-up CT scan/MRI obtained at 12 months demonstrated durability of the spinal reconstruction, in spite of the fact that possibility of high recurrence following the surgery and radiotherapy (Fig. 2a–d).

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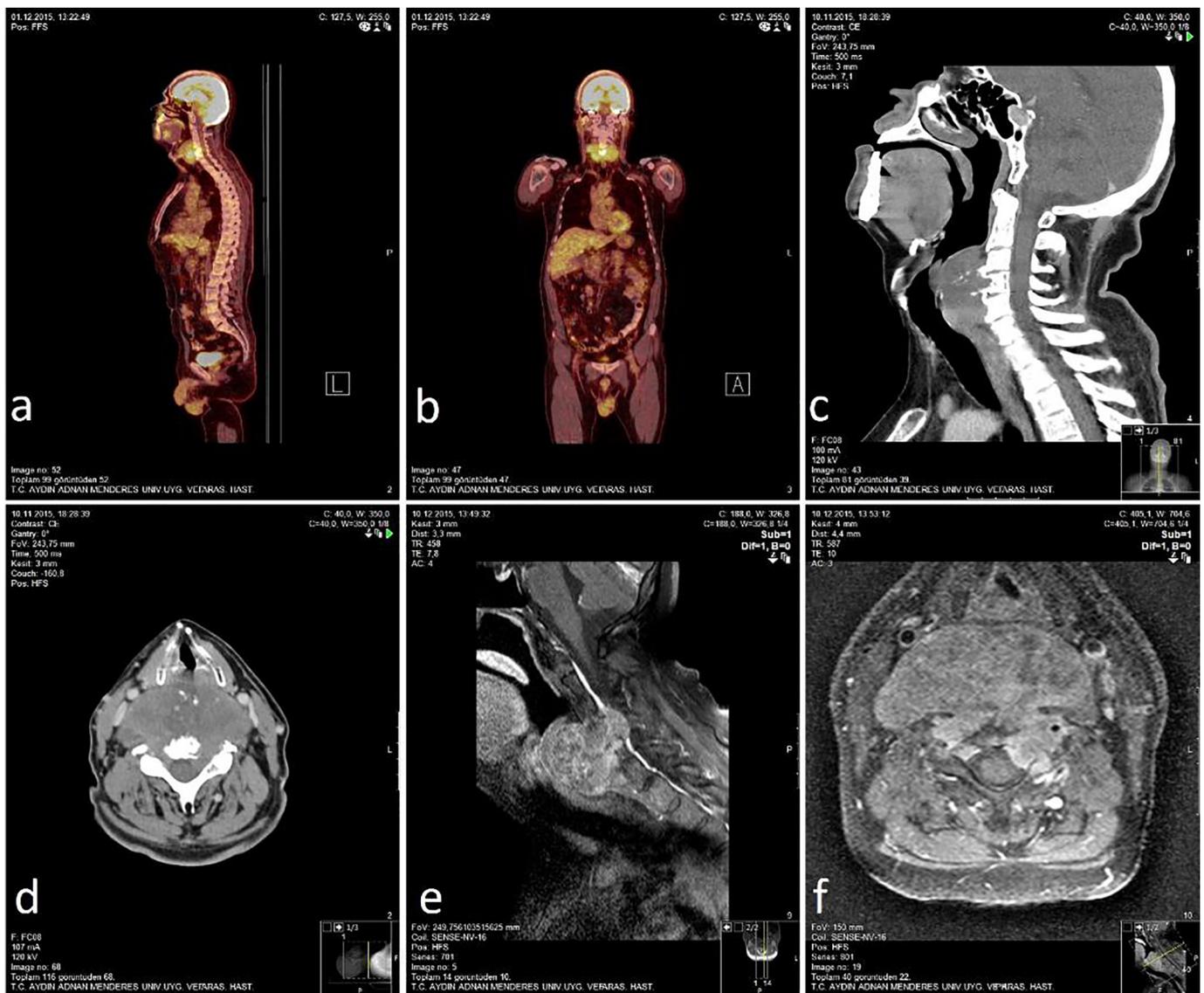


Fig. 1. a, b A whole-body [¹⁸F]-fluorodeoxyglucose positron emission tomography-CT scans showed a hypermetabolic large lesion extending from C2 to C6 in the cervical region with evidence of destruction of C4 vertebral body. c, d CT scan of the cervical spine revealed the prevertebral mass lesion destructing the C4 vertebra. e, f MRI of the cervical spine confirmed the prevertebral mass lesion with heterogenous intensity compressing both cervical spinal cord and prevertebral structures including esophagus, trachea, and vertebral arteries.

3. Discussion

To our knowledge, this is the first case of cervical spine chordoma diagnosed as OSA at the first in the current literature, possibly due to simply poor investigation of the case despite having neurological finding in his upper extremity, namely numbness. It is well-known that there is a primary form of OSA which has several proposed aetiologies and a secondary form of the disease such as nasopharyngeal tumour such as carcinoma or in this instance chordoma involving the cervical vertebra. Once the histological diagnosis is available, it has been demonstrated that one- or two-stage spondylectomy by a multidisciplinary team than subtotal resection is ideal for resection of chordoma with less recurrence and no metastasis, but this can be difficult in the cervical spine due to a higher rate of complications of this approach, in particular in the presence of the involvement of multiple levels of the nerve roots and vertebral arteries [1,2]. Therefore, spondylectomy is technically challenging in the cervical spine and very few reports of cervical spine spondylectomy for chordoma exist in the literature [2–5]. Barrenechea et al. [6] also reported their series of 7

patients with cervical spine chordoma who were treated using piecemeal resection because of treatment difficulty in chordoma of the cervical spine. In the current case, we have used a subtotal resection technique, called corpectomy, as an alternative to spondylectomy for removal of chordoma in the cervical spine with preservation of neighbouring critical nerve roots and the vertebral arteries involved. It has been shown that radiotherapy, following resection of the tumour with fusion by an iliac crest graft and plate stabilisation, is effective in the long-term control of these aggressive tumours [2,7]. In our case, however, we preferred to perform only anterior corpectomy because combined posterior and anterior approaches have higher associated morbidities and to do it as staged operation, if necessary in the future, to allow the patient some time to recover. Now, we believe that our strategy is useful for such cases because of its low morbidity rate and its simplicity compared to spondylectomy.

In patients presenting with symptoms of OSA secondary to cervical spine chordoma and/or multinodular goitre, an increased awareness and imaging of the cervical spine with CT and/or MRI might avoid unnecessary prescription of CPAP or BiPAP machine in such cases.

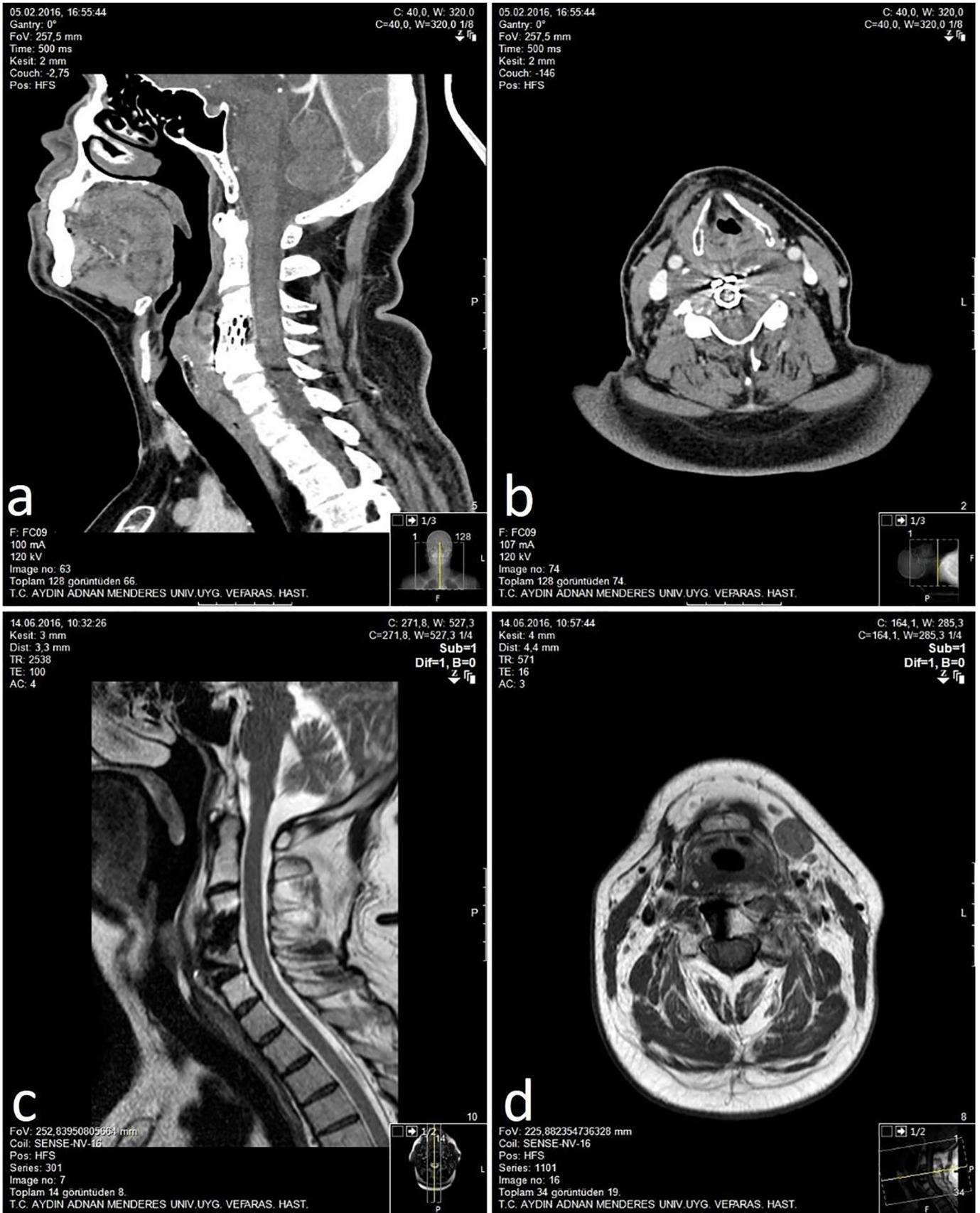


Fig. 2. a–d A follow-up CT scan/MRI obtained at 12 months demonstrated durability of the spinal reconstruction.

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Conflict of interest

The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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