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C2 Neurilemmoma: Originally Misapprehended as Psychogenic Headache and Nuchalgia — Case Report

Chui-Lee Wong, Mei-Hue Hsu

A patient with headache and nuchalgia for three years was initially diagnosed as psychogenic origin. Spinal cord compression syndrome was unrecognized until motor evoked potential study showing abnormal findings. Excision of a nearly 3×2×2 cm dumbbell extra-and intradural tumor, which arose from the right C2 root, was performed after MRI documentation. Histologic diagnosis was neurilemmoma. Motor evoked potential study is useful in spinal cord diagnostics. The particular value of complete history-taking and exact neurological examination should not be underestimated.

Key words: neurilemmoma, cervical root, motor evoked potential

INTRODUCTION

The most common causes of headache and nuchalgia meet at physical medicine and rehabilitation clinic are cervical spondylosis, postural strain and psychogenic origin. However, headache and nuchalgia are the most important and earliest symptoms occurred in patients with upper cervical neurilemmoma [1], who are rarely referred to a physiatrist before diagnosis and surgical removal of the tumor indeed. This report presents a patient with C2 neurilemmoma, whose root symptom was originally misapprehended as psychogenic headache and nuchalgia, and the spinal cord compression syndrome was recognized only after motor evoked potential study showing abnormal findings.

CAAE REPORT

A 39 year-old female was referred to the clinic because of pain over the occipital area, nuchalgia and rigidity of the neck for 3 years. After heat therapy for five days, the patient complained of the heat therapy was ineffective to relieve her neck pain and headache. Besides, she emphasized the need of straining on voiding for one and a half years, constipation and slight weakness of her left arm for one month. Mild hyperreflexia of all limbs,transient clonus of the left ankle, mild hypesthesia to pin prick over entire body up to C3 and slight weakness of the left arm were recognized as long tract signs only after motor evoked potential study showing abnormal findings (Table 1). The cortex latency and the central motor latency were mildly prolonged on the left side. The side to side differences of cortex latency and central motor latency were significantly abnormal. The motor evoked potential study suggested corticospinal tract dysfunction between the right motor cortex and the low cervical spine.

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Table 1. Latencies (mSec) of MEPs to cortex and C7 root stimulation, recording from abductor digiti minimi.

	Cortex Latency (CL)	Root Latency (RL)	Central Motor Latency (CML)
Left ADM	21.8	13.1	8.7
Right ADM	18.2	11.8	6.4
Side to Side Difference	3.6	1.3	2.3

CML: difference between CL and RL; ADM: abductor digiti minimi.

Plain films of the cervical spine were normal. Spinal CT showed a large soft tissue density tumor at right C1-2 level with compression on the cord (Fig. 1). MRI revealed a nearly 3×2×2 cm dumbbell hyperintense extramedullary tumor at C1-2 level crossing over the markedly dilated right C1-2 neuroforamen with compression over the cord ventrally on the T2*W image. The lesion showed marked contrast enhancement on Gadolinium-T1W image (Fig. 2). A laminectomy was carried out and an extra- and intradural tumor, which arose from the right C2 root, was excised. Histologic diagnosis was neurilemmoma (Fig. 3).

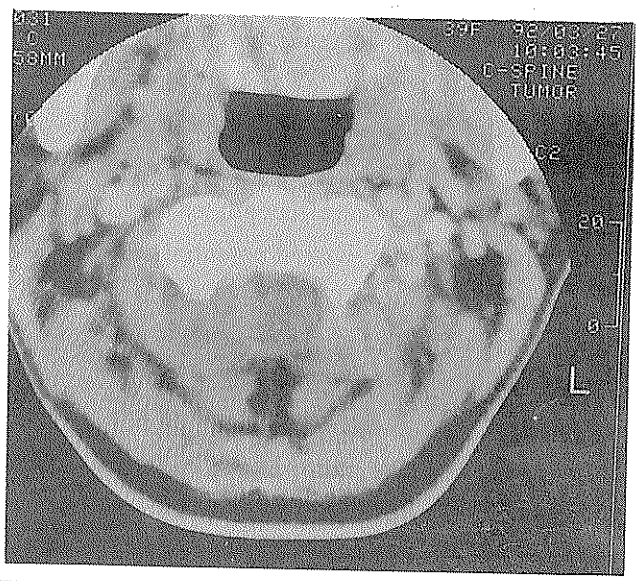

Fig. 1. Spinal CT with contrast enhancement showing a large soft tissue mass (arrowheads) at the right C1-2 level with compression on the cord.

One week after surgery, symptoms and signs disappeared except slight hyperreflexia of the upper limbs. The follow-up MRI, which was performed eight months after surgery, revealed good recovery of the previously compressed cervical cord and no recurrence of tumor (Fig. 4).

DISCUSSION

The prevalance of neurilemmomas among space-occupying spinal tumor has ranged from 11% to 38%. In 1972, Nittner reviewed 4885 cases of space-occupying lesions published since 1940 and found 23.1% of these lesions were neurilemmomas. Among these spinal neurilemmomas, 71.8% were intradural and extramedullary, 12.8% extra-and intradural, 14.1% extradural, and 1.3% intramedullary [1]. A few cases of intraosseous neurilemmoma of spine has been reported [2]. Tonnis found 29% of spinal neurilemmomas to be cervical, 59% thoracic and 12% to be lumbar. Broager noted the mean age of development of symptoms and the mean age at surgery of cervical neurilemmomas was 34 and 40 years, respectively. This gave a mean length of history of 6 years. The neurological symptoms of patients with spinal neurilemmomas are due primary to spinal nerve root disturbance and secondarily to spinal cord compression. Pain, which is present in 85% of patients, almost always is the first and most important symptom. In upper cervical

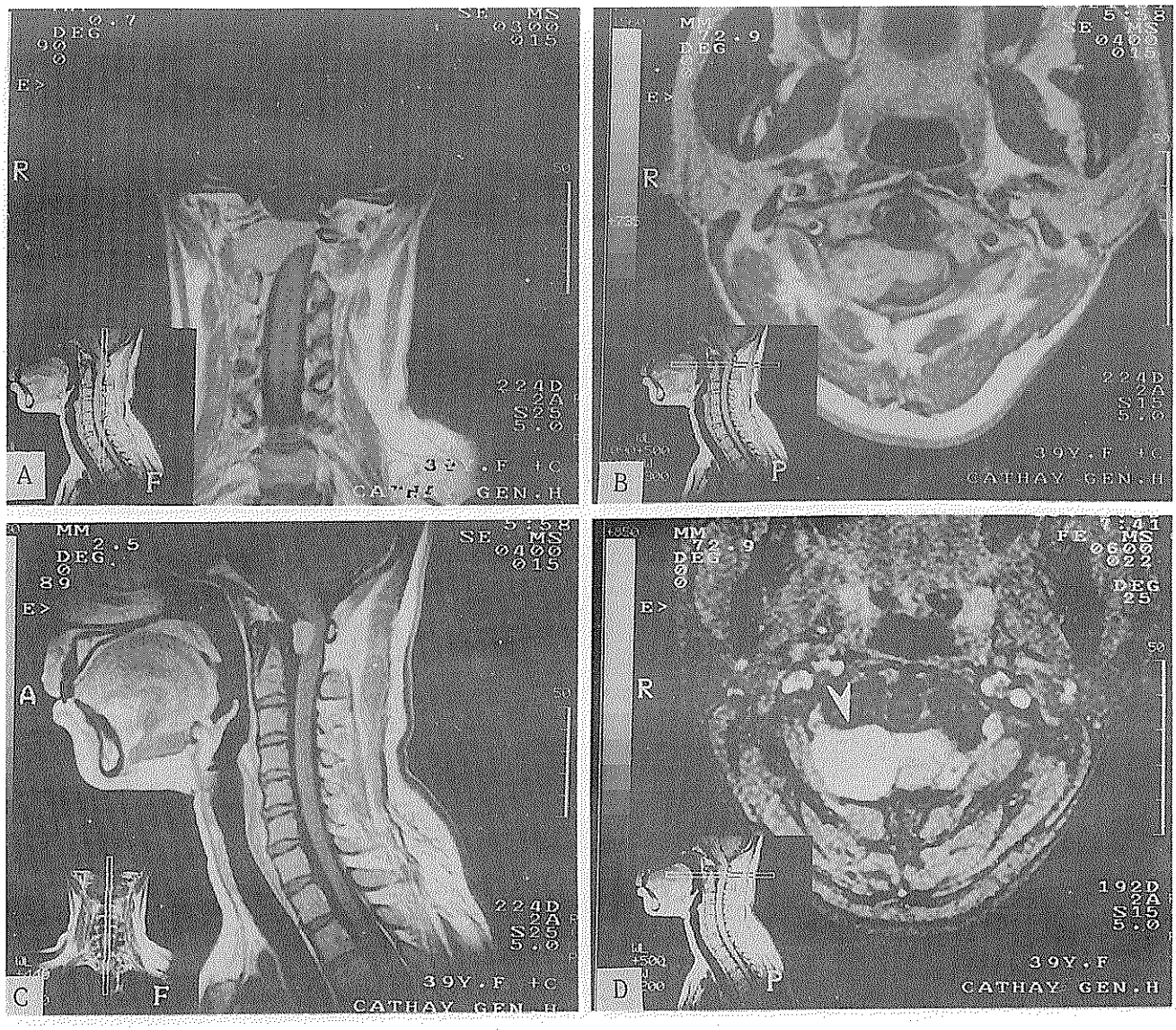

Fig. 2. MRI coronal (A), axial (B), and sagittal (C) GD-T1W images revealing a nearly 3×2×2 cm dumbbell extramedullary tumor crossed over the markedly dilated right C1-2 neuroforamen with compression over the cord ventrally. The tumor (arrowhead) is hyperintense on axial T2*W image (D).

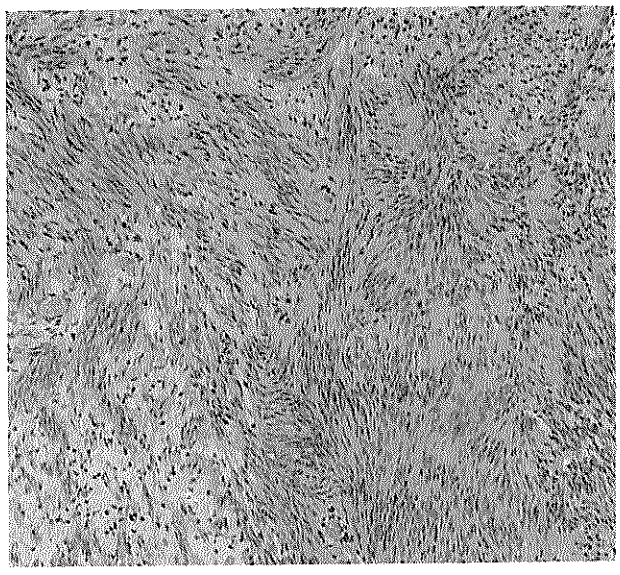

Fig. 3. Photomicrograph of the tumor showing pallisading nuclei (H & E, x150).

neurilemmoma, pain is mainly distributed in the posterior neck or the occipital area. About 85% of patient with spinal neurilemmomas had disturbance of motility during the course of the disease. Of those who showed no motor disturbance, half had reflex abnormalities. Hyperreflexia with retention of normal power is a typical finding in neurilemmomas of the cervical and upper thoracic segments. About

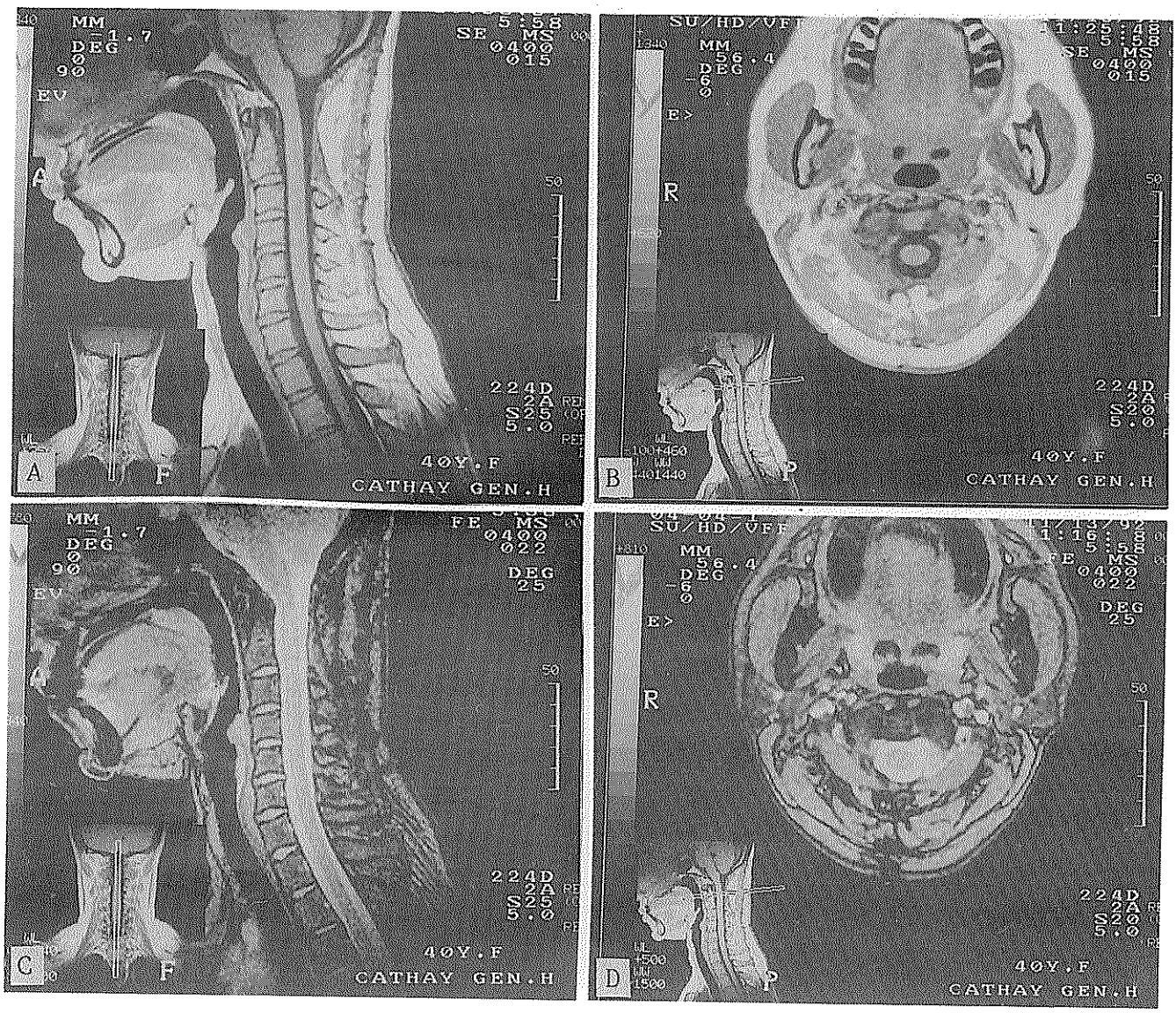

Fig. 4. Postoperative MRI showing no recurrence of the tumor and good recovery of previously compressed cervical cord (A, sagittal, and B, axial T1W images; C, sagittal, and D, axial T2*W images).

half of the patients with cervical neurilemmoma had bowel and bladder dysfunction [1]. Increased intracranial pressure, papilledema, dementia [1,3] and subarachnoid hemorrhage [4] associated with spinal neurilemmomas has been noted. The etiology of increased intracranial pressure is unknown. It is probably caused by defective CSF reabsorption secondary to an elevated protein level and episodes of clinical or subclinical subarachnoid hemorrhage [3]. Local compromised circulation of branches of the vertebral artery arises when the neurilemmoma extends beyond the upper level of C2 and into the posterior cranial fossa [1,5]. Other rare clinical manifestations in high cervical neurilemmomas are astereognosis and amyotrophy of the hand [6], and trigeminal neuralgia [7] associated with other symptoms and signs of cord compression syndrome.

In cervical neurilemmomas, the mean duration of the first symptom before the second appears is 2.8 years. Since spinal neurilemmomas characteristically show a long interval between the onset of the first symptom and the development of further

symptoms or signs, only 15% of these patients are diagnosed at early stage [1]. Before MRI, spinal neurilemmomas were mainly revealed by myelography and/or CT myelography. It needs appropriate techniques and runs higher risk to demonstrate and localize the high cervical neurilemmoma in myelography. MRI is better in demonstrating their relationship with and mass effect on the adjacent spinal cord. Neurilemmomas are markedly hyperintense compared with the spinal cord on T2W images. They are also well demonstrated on TlW images with gadolinium enhancement [8]. Preoperative vertebral angiography is advised when the vertebral artery may be compromised by a large cervical neurilemmoma or a upper cervical neurilemmoma with extension into the posterior cranial fossa [5,9].

There is good functional recovery after excision of the spinal neurilemmomas [1,10]. The risk of causing disabling neurological deficit after sacrificing a root critical for the function of the upper or the lower limbs is small [11].

In the present case, the long interval between the appearance of each relatively mild symptoms, and the equivocal neurological signs distracted the physicians from considering each symptom to be correlated with a space-occupying spinal lesion, and misled into the impression of psychogenic origin until motor evoked potentials to magnetic stimulation showing abnormal findings. Magnetic stimulation of the motor system enables examination of the central motor pathways in awake subjects. This noninvasive neurophysiologic technique is useful in spinal cord diagnostics [12]. It is of meaningness as an less expensive, preliminary referable diagnostic method when encountering symptoms which led to the suspicion of space-occupying spinal lesion but there is no definite neurological signs yet.

Retrospectively, the present case described the nature of headache and nuchalgia as becoming persistent and exaggerating at mid-night three months prior to CT and MRI documentation of the lesion. The pain, as such, implies organic cause. So the particular value of thorough history taking and exact neurological examination should not be underestimated in dealing with these problems. There are far more real pathologic headache problems which are at first thought to be psychogenic than vice versa. The diagnosis of psychogenic headache should not be made impudently before all physical causes of the pain have been ruled out [13].

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第二頸神經鞘瘤——誤為心因性頭頸痛～病例報告

黃翠莉　許美慧

本篇報告一位因未被發覺為頸神經鞘瘤導致頭頸痛而轉介至復健科的39歲女性病例。其三年病史中先後出現的症狀有後頸和後腦部疼痛、頸部僵硬、解小便需用力，均被誤以為心因性的問題。復健五天後，患者強調便秘一個月，左上肢稍乏力，熱療無法減緩頭頸痛。在Motor Evoked Potential測驗顯示異常後，始確認其具脊髓受壓迫症候群。磁振影像掃描發現在第一、二頸椎處右方有一個3×2×2公分，啞鈴形的腫瘤將脊髓往左、後方擠壓。手術發現此瘤出自右側第二頸神經根，其病理為神經鞘瘤。經手術切除一週後，上述症狀消失。

疼痛是此類患者最常發生的第一個症狀，高位頸神經鞘瘤的疼痛主要位在後頸及後腦部，疼痛與以後出現症狀的平均間隔時間為2.8年。故只有較少患者被早期正確診斷。從本例的診斷過程亦得悉脊髓受壓迫症候群仍未確定但又可疑時，Motor Evoked Potential測驗不失為一個可供參考的初步檢查工具，並以斷層掃描及磁振影像掃描證實診斷，而後手術切除腫瘤。

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