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Metastatic Tumor in the Atlantoaxial Spine: A Case Report

Hsin-Chi Wu, Chieh-Tsai Chiu, Pu-Sheng Yeh, Yi-Shiung Horng

Departments of Physical Medicine and Rehabilitation, Neurosurgery, and Chest Medicine, En Chu Kong Hospital, Taipei.

Atlantoaxial involvement is rare in metastatic tumors of the spine. We report a case of atlantoaxial metastatic adenocarcinoma of pulmonary origin, which was treated as cervical spondylosis because the first clinical manifestations were cervical pain followed by limitation of rotation. In the literature, most authors stated that in patients with cervical spine metastases but without neurological signs, pain may be the only complaint. Most metastasized tumors of the atlantoaxial spine originate in the lung or breast. The goal of treatment for these cases is palliative. Posterior fixation is indicated for patients having intractable pain or whose cervical spine is unstable. Conservative treatment for the rest of their lives is recommended for these patients. (Tw J Phys Med Rehabil 2006; 34(2): 97 - 102)

Key words: cervical metastasis, cervical pain, atlantoaxial spine

INTRODUCTION

Metastatic cervical malignancy is not common in patients with cervical pain. A careful clinical history may arouse suspicion of a metastatic tumor of the atlantoaxial spine. Magnetic resonance imaging (MRI) and computed tomography (CT) examinations are helpful in arriving at a diagnosis. In view of the poor prognosis, conservative treatment, either external beam radiation therapy or chemotherapy with external support, is recommended for those patients with a stable spine. Surgery is indicated for patients whose spine is unstable. This report presents a case that was initially misdiagnosed as ordinary cervical pain. One month later, a cervical malignancy was discovered.

CASE REPORT

An 83-year-old man was admitted to our hospital on July 26, 2002. His chief complaint was a cough of two months duration. This was consistent with his past history of pneumoconiosis. This patient also had a history of cervical spondylosis that had been diagnosed at a medical center and treated with cervical traction for about one month prior the first admission.

First Admission

Right pneumothorax was detected when he visited our emergency room (ER). A chest tube was inserted and his condition improved greatly. The tube was removed four days later.
His doctor consulted the department of Physical Medicine and Rehabilitation (PM&R) for his neck pain on the day the chest tube was removed. Limitation of range of motion of neck was about terminal 30 degrees in bilateral lateral rotation and hypoesthesia was present from occiput to vertex. A C1-2 open mouth radiograph was taken. A previous cervical spine plain film taken at a medical center showed spur formation in the cervical spine. Neurological examinations, including muscle power and deep tendon reflexes of four limbs, were normal. Therefore, we did not recommend any rehabilitation therapy until we were able to confirm a diagnosis. The patient was discharged on August 1, 2002 after a C1-2 open mouth film was taken without follow-up of the result of the film. After his discharge, the patient received a local injection and cervical traction at the medical center located close to his home.

**Second Admission**

The patient revisited our PM&R clinic on August 8, 2002 because of persistent symptoms. He was immediately transferred from the PM&R clinic to the neurosurgery clinic because of C1-2 subluxation and blurred odontoid process on the C1-2 open mouth plain film and C spine plain film (Figure 1). He was readmitted with suspicion of a C2 lesion. We performed CT of the cervical spine on August 9, 2002 (Figure 2). This showed C1-2 subluxation associated with an osteolytic lesion in C2 and a periarticular soft tissue mass. The results of MRI (Figure 3) performed the same day, revealed C1-2 subluxation complicated by left vertebral artery thrombosis and PICA territory infarction and a C2 mass.

Chest CT (August 14, 2002) (Figure 4) showed minimal pneumothorax, pneumoconiosis with fibrotic change and subpleural bullae formation, and RLL mass at the superior segment.

On August 14, 2002, surgery for C1-2 fixation was performed. At the same time, trans-oral biopsy of the C2 mass was performed by our ENT physician and the subsequent pathology report confirmed metastatic adenocarcinoma. The patient was referred to the National Taiwan University Hospital (NTUH) for chemotherapy and radiotherapy on August 30, 2002. From September 3 to late September, the patient received radiotherapy over C1-C7 with 3000cGy/10 fraction and oradexon use at NTUH.

The patient developed gait disturbance and left side dysmetria on September 13. CT examination suggested cerebellar infarction. The patient received chemotherapy with Gemcitabin 1200mg on September 24 after his brain had stabilized.

Unfortunately, as a complication of chemotherapy, the patient developed leucopenia and pneumonia. He was transferred to the intensive care unit (ICU) because of septic shock and suppurative parotiditis on September 30. He returned to the step-down unit on October 6 after sepsis was under control. After control of suppurative parotiditis, the patient went home on November 19.

**Third Admission**

The patient was readmitted to our hospital on December 4 suffering from pneumonia with respiratory failure and septic shock with complication of febrile neutropenia following chemotherapy. Sputum culture found MRSA and pseudomonas. He returned home to be cared by his family and died of multiple organ failure and infection on March 1, 2005.
After reviewing the patient’s history, we found he had no neurological complaint with the exception of nuchal soreness and mild stiffness on his first visit to a medical center. Subsequently, he developed occipital
numbness and ROM limitation in lateral rotation. Neither neurological deficits nor long tract signs was found before or after the operation except occipital numbness.

**DISCUSSION**

Degenerative disease of the cervical spine is the most common cause of neck pain in the elderly. Increased incidence of cervical spondylosis, herniated disc, tension nuchalgia or fibromyalgia. Contraindications for cervical traction are tumors, an unstable spine, and severe osteoporosis. The key to developing a successful treatment plan is making a careful differential diagnosis. Careful physical examinations, frequent follow-ups of the effects of therapy and MRI examinations help the physician decide on the right course of treatment.

**Carcinoma with Spinal Metastasis**

About 5% to 10% of patients who are diagnosed with cancers have spinal metastases, predominantly thoracic spine, followed by lumbar spine and cervical spine. However, symptomatic metastases of the atlantoaxial spine are rare. The character of tumor which metastasis to the cervical spine kept highly variable. Jonsson et al mentioned the most common origins of cervical spine metastasis were breast cancer and myeloma. However, Roa’s study showed prostate, breast, and lung neoplasms accounted for 57% of the cervical metastases. Histological diagnoses also varied widely in cancers with atlantoaxial metastases. Lung tumor, breast tumor, prostate tumor and sporadic case reports of von Recklinghausen neurofibromatosis, nasopharyngeal rhabdomyosarcoma, colon cancer, pituitary adenocarcinoma, esophageal adenocarcinoma, and thyroid carcinoma have all been found in the literature. Associated nonspinal skeletal, extraskeletal, or multiple-level spinal metastases were seen in most patients.

**Clinical Presentation of Atlantoaxial Tumor**

A careful clinical history and physical examination combined with a high degree of suspicion suggest a metastatic tumor to the atlantoaxial spine. Cervical pain as the only complaint in a patient with metastatic advanced lung carcinoma was reported by Bobek et al. Mechanical neck pain elicited in flexion, extension, and lateral rotation was noted. Pain on lateral rotation can distinguish an atlantoaxial tumor from a tumor occurring in the subaxial cervical spine. In cases with atlantoaxial spine involvement, the quality and intensity of the pain does not distinguish patients with normal spine alignment from those with significant fracture subluxation. Severe mechanical pain does not necessarily indicate spinal instability that requires surgery correction. Rao and colleagues reported that pain is the initial symptom in 89% of 21 cases and no patient with cervical spine metastasis experienced neurological deficits. Three cases (16%) had slight radiographic collapse and deformity. However, only one (5%) patient had documented instability of the cervical spine. Neurological deficits are rare. Occipital neuralgia usually occurs in patients with C2 nerve root involvement. The prognosis for these patients was poor. Mean survival after diagnosis was 5 to 14.7 months.

We urge an early recognition of the aforementioned symptoms and the use of CT and MRI to improve the detection of tumors in the atlantoaxial spine before fracture subluxation occurs.

**Treatment in Cervical Metastasis**

Patients with normal alignment or minimal subluxation were considered for nonoperative therapy, either external beam radiation therapy or chemotherapy with external support combined with high dose of steroids. Surgery was reserved for patients with significant fracture subluxation, including atlantoaxial displacement of more than 5mm or exceeding 11 degrees in angulation with displacement more than 3.5 mm. An anterior approach and excision of the metastasized tumor was advocated by some authors. However, most authors have not found this to be necessary. Posterior stabilization of the spine in order to avoid poorly tolerated external orthoses, is the best choice for patients with severe nuchal pain and atlantoaxial instability. In these articles, investigators reported that most of the patients who received posterior stabilization had pain relief and none of these patients had problems of neurological decline or sudden death during the follow-up.
period. Posterior occipitocervical stabilization was chosen when the odontoid process was totally destroyed.\[13\] In recent years, an increasing number of posterior approach procedures with odontoid screw and C1-2 transarticular screw fixation have been performed.\[1,15,16\] However, some authors used cervical vertebroplasty to treat an osteolytic lesion of the atlantoaxial spine. They injected cement to the upper cervical spine via a posterior percutaneous or a transoral approach, under fluoroscopic guidance. These authors claimed that it is a safe and effective method of surgically treating patients.\[17-19\] If possible complications, such as infection and extravasations of cement could be controlled; vertebroplasty may be the treatment of choice for a potentially difficult atlantoaxial spine metastatic tumor in the future.

**CONCLUSION**

The clinical diagnosis of cervical spine malignant metastasis is based on taking a detailed history and on a thorough physical examination. MRI and CT are helpful in arriving at a differential diagnosis. Neck pain is usually the only complaint in the atlantoaxial spine metastatic tumor. This paper and extensive reports in the literature all recommend conservative treatment for these patients for the rest of their lives. Posterior fixation or vertebroplasty is recommended for patients with an unstable spine.

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寰樞椎恶性腫瘤轉移：病例報告

吳欣治  邱建才¹  葉步盛²  洪怡珣

財團法人恩主公醫院 復健科 神經外科¹  胸腔科²

頸椎惡性轉移腫瘤是頸椎疼痛鑑別診斷之一。在此病例報告中，病患因頸部疼痛早先以頸椎間關節炎於某一醫學中心接受頸椎牽引，後因氣胸住進本院胸腔科病房。在復健科照會中，理學檢查發現頸椎旋轉活動度降低、後腦麻感，懷疑第一二頸椎問題而接受以下檢查。X-ray 檢查顯示頸椎第一二節滑脫，後續之電腦斷層及磁振造影檢查證實頸椎第二節腫瘤。在轉移性頸椎腫瘤的診斷下接受頸部固定手術，化學治療及放射治療。在文獻中顯示，頸椎腫瘤通常只以頸部疼痛為唯一症狀，出現神經症狀者少；主要的轉移源為肺癌及乳癌，治療則以保守治療為主。頸椎疼痛及不穩(unstable or subluxation)時接受後側固定手術(posterior fixation)。（台灣復健醫誌 2006；34(2)：97 - 102）

關鍵詞：脊椎轉移性腫瘤(cervical metastasis)，頸痛(cervical pain)，寰樞椎(atlantoaxial spine)