

Incidence of Motor Neuron Disease (MND) in Patients Diagnosed with Cervical Spondylotic Myelopathy (CSM)

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ABSTRACT

Objective: Motor neuron disease (MND) can occur in patients with cervical spondylotic myelopathy. The purpose of this study was to determine the prevalence of motor neuron disease in individuals suffering from cervical spondylotic myelopathy (CSM).

Materials & Methods: A prospective study was conducted at the Lady Reading Hospital. The patients (n = 70) included were between ages 55 to 75 with established clinical and radiological diagnoses of CSM. Motor neuron disease was diagnosed clinically by Awaji criteria. The patients were evaluated with NCS/EMG (Nerve Conduction Studies/Electromyography) and followed for 6 months. The motor neuron disease was confirmed by modified El Escorial criteria.

Results: 40 patients had bilateral upper limb weakness (57%), however, the rest of the 30 patients had unilateral upper extremity weakness (42.8%). Hoffman's sign was positive in 60% of patients. Lhermitte's sign was also positive in 31% of patients. The majority of patients (55.7%) were found with grade 4 for deltoid followed by 51.4% patients for biceps brachii. MRI showed anterior cord compression at the level of C5 and C6 in 27.14% of patients and C6 C7 in 32.8% of patients. 15.7% of patients were suspected of having both CSM and MND. The incidence of MND in our study was 5.7%. Nurick grade III was reported in 45.7%, grade IV in 24.2%, grade V in 14.2%, and grades I & II in 7.14% and 4.2% of patients, respectively.

Conclusion: CSM and MND can occur in the same patients. The incidence of MND is 5.7% in patients with cervical spondylotic myelopathy, hence in CSM patients, workup must be done for MND as it affects the outcome of surgery in these patients.

Keywords: Cervical Spondylotic Myelopathy (CSM), Motor Neuron Disease (MND), Awaji Criteria, NCS/EMG (Nerve Conduction Studies/Electromyography), El Escorial Criteria, ALS (Amyotrophic Lateral Sclerosis), Muscle Strength.

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INTRODUCTION

This study sought to determine the prevalence of motor neuron disease in individuals suffering from cervical spondylotic myelopathy. Patients with cervical spondylotic myelopathy present with difficulty in walking, upper limb weakness, and neck pain. As the world's population ages, cervical spondylotic myelopathy (CSM), the primary cause of acquired spinal cord dysfunction, is predicted to become more common. Although the clinical characteristics of CSM can vary greatly, chiropractors commonly treat patients who exhibit similar CSM symptoms such as neck discomfort, weakness in the extremities, and abnormal gait. Early detection of myelopathy-related symptoms may reduce future disability and enhance quality of life.¹ The use of electrophysiologic testing aids in ruling out other possible diagnoses. It is unknown if conservative therapies are beneficial. While there is a risk involved, surgical decompression helps some patients' neurologic function and keeps it from getting worse in others. Neurologists must be knowledgeable about this widespread ailment. Patients who have minor CSM symptoms and signs might be closely watched. Patients with progressive, moderate-to-severe neurologic impairments should be evaluated for surgical decompression using either an anterior or posterior route. To rule out other etiologies and assess the existence and severity of myelopathic alterations in a patient presenting with apparent cervical myeloradiculopathy, a thorough clinical examination with ancillary testing, such as neuroimaging and electrophysiologic investigations, is crucial. For many patients with mild CSM, conservative medical therapy under close supervision is adequate. Surgical intervention should be considered for individuals who have substantial neurologic impairments that are getting worse. Younger age, shorter illness duration, and milder neuroimaging abnormalities

may be predictive variables for a positive surgical outcome; however, these results are not consistent across studies, and further study is required.²

Ferguson and Caplan (1985)³ divided cervical spondylotic myelopathy into four clinical syndromes which included radiculopathy only, radiculopathy and myelopathy, pure myelopathy with no radicular symptoms, and vascular involvement. The most common form is combined radicular and myelopathy symptoms. There are radicular symptoms at the level of cervical cord compression and upper motor neuron findings below the level of the lesion. According to Simeone & Rothman (1982),⁴ it is the most common cause of spinal cord dysfunction above the age of fifty-five years characterized by stooped posture and wide-based jerky gate. The reflexes are diminished at the level of compression however they are exaggerated at the level below the compression. There will be diminished biceps reflexes at the level of C5 C6 compression whereas the triceps will be exaggerated. Also, there will be a positive Hoffman's sign indicating the upper motor neuron involvement below the level of compression. If the C6 root is compressed, there may be a decreased brachioradialis reflex, which is linked to findings of upper neuron lesion (UMN) in finger flexion. The sensation may be reduced below the level of CSM. Dorsal root compression will lead to dermatomal sensory loss and posterior column compression will result in reduced position and vibration on the same side of involvement however, spinothalamic tract dysfunction will lead to opposite side pain and temperature loss. Involvement of the bladder is an uncommon manifestation. An electric shock-like feeling experienced when flexion or extension of the neck occurs in the lower extremities may be a positive characteristic of Lhermitte's sign. According to Edwards et al, (1985),⁵ there may be combined spondylosis in both cervical and lumbar regions in 13% of cases and surgeons

prefer to manage cervical spondylosis first. In these cases, there is a mixed clinical picture of UMN findings in lower extremities with diminished ankle reflex. Gait abnormality may be assessed by using the NURICK grading scale⁶⁻⁸. The mJOA (modified Japanese Orthopedic Association) scale can be used to assess the severity of myelopathy. Its components include motor power in arms/legs, sensations in arms/legs, and bladder function. This 18-point investigator-administered scale covers sensory loss of the upper extremity, sphincter dysfunction, and motor dysfunction of the lower extremities, respectively.⁹⁻¹⁰

Cervical spondylotic myelopathy must be distinguished from other clinical entities like motor neuron disease which may have a similar clinical picture. Motor neuron disease is sporadic (90-95%), however, some familial cases have been reported as well (5-10%). Diagnostic criteria for MND were suggested at the World Federation of Neurology conference at Al-Escorial (Spain 1990)¹¹⁻¹³. Motor neuron disease (MND) has sporadic onset and it presents with muscle wasting, fasciculations, and emotional lability. It is seen in 2 per 100000 population yearly and prevalence is 4-6 per 100000 population. Men are affected more than women 1.5:1¹⁴⁻¹⁶. Any sensory signs and symptoms are absent and there may be mixed findings of upper and lower motor neuron involvement. The disease is common in the 6th and 7th decades and there is often asymmetrical presentation however 5% of cases may be in younger age groups due to familial involvement risk factors include toxins and farming. Motor neuron disease patients have bulbar symptoms in 25% of cases.¹⁷⁻¹⁸ Like dysphagia, tongue weakness and fasciculation, dysarthria slurring of speech. Earliest symptoms include asymmetrical weakness of the upper extremities. There is distal weakness of extremities evident by difficulty in opening bottle tops, and gripping different objects. There is involvement of arms in 35% of cases. Muscle fasciculation may precede

weakness by several months. There may be combined upper and lower motor neuron involvement in 65% of cases at presentation¹⁹. Investigations include MRI of the brain, spine, and EMG. The following EMG parameters are considered when diagnosing motor neuron disease: (i) There is fibrillation and fasciculation in the muscles of the head, the lower and upper limbs, or both. In more severely affected muscle nerves, they are at least 70% of the average normal value according to age; (ii) there is a reduction in the number of motor unit action potentials and an increase in their amplitude and duration; (iii) normal electrical excitability of remaining motor nerve fibers and motor fiber conduction velocity; and (iv) normal excitability and conduction velocity of sensory nerve fibers are present even in severely affected extremities. MRI excludes CSM, and neoplasm and also shows enhanced T2WI signals in white matter tracts, spinal cord brainstem, and internal capsule.²⁰⁻²¹ Following four randomized clinical studies with 1477 patients, Rivuzole therapy is currently the standard of care for MND patients and is backed by excellent recommendations in the UK. The median survival time after the beginning of symptoms is 3.5 years, and non-invasive ventilation has been demonstrated to both extend life and enhance quality of life. Poor prognostic indicators are female sex and bulbar onset, age < 50 years have prolonged survival than onset at age > 50 years.²²⁻²³

MATERIALS AND METHODS

Study Design

A prospective, observational study was conducted at the Neurosurgery Unit of Lady Reading Hospital from 1st February 2021 to 30th May 2022.

Inclusion Criteria

In all, 70 individuals with a diagnosis of cervical spondylotic myelopathy were included in this

investigation. Every patient in the group had symptoms and signs of cervical spondylotic myelopathy (CSM) and was equal to or older than 55. After receiving approval from the ethical review board and with the consent of the patients, an MRI was used to make the diagnosis.

Exclusion Criteria

Patients below 55 years of age and those having a history of trauma or previous surgery for the cervical spine were excluded from this study. Patients having co-morbid like diabetes, and hypertension were also excluded from this study.

Clinical Assessment Tools

Data was collected on a proforma that contained the relevant information including patient age, gender, duration of signs and symptoms, MRI, and NCS /EMG findings. We used the Nurick grading scale (from grade 0 to VI; 0=no root/cord symptom; I=symptom of root involvement only; II=signs of spinal cord/normal gait; III=mild gait abnormality; IV= moderate gait abnormality; V=walk with assistance & VI= chair bound/bedridden) and MRC (Muscle Research Council for the assessment of muscle strength) scale (0-5) to assess functional status of the patients. All the patients were confirmed by MRI findings of CSM and they were subjected to NCS/EMG. Awaji criteria were used for clinically probable MND and all the patients were followed for 6 months and diagnosis was confirmed by modified El Escorial criteria (Suspected ALS, Possible ALS & Probable ALS).

RESULTS

Gender Incidence

Out of 70 patients, 44 were males (62.8%) and 26

were females (37.14%).

Age Incidence

The patients included in our study were from 55 years to 75 years with a mean age of 65 years.

Signs & Symptoms Reported

Out of 70, 40 patients had bilateral upper limb weakness (57%), however, the rest of the 30 patients had unilateral upper extremity weakness (42.8%). In the latter group, 22 patients had right-sided weakness (73%) as compared to the left side (26.6%). Hoffman's sign was positive in 42 patients (60%) out of 70. Lhermitte's sign was also positive in 22 (31%) patients in our study. Bulbar signs and symptoms (tongue fasciculation/dysphagia/ dysarthria) were observed in 3 patients (4.2%). Weakness was more common in the distal muscle group as compared to proximal muscle groups.

Muscle Strength from the MRC Scale

Muscle power was assessed in individual muscle groups using the MRC scale (from 0-5) (Table 1). According to the assessments, the majority of patients (55.7%) were found with grade 4 for deltoid followed by 51.4% of patients for biceps brachii. The majority of patients (50%) were included in grade 3 for triceps and 45.7% of patients in grade 2 for wrist extensors. 37.1% of patients included in grade 1 for finger flexors. Only 1.4% (for wrist extensors) and 14.2% (for finger flexors) were categorized into grade 0. Only 17.1% were categorized into grade 5 for deltoid and biceps brachii, 2.8% for wrist extensors, 1.4% for finger flexors, and 10% for triceps. Details are mentioned in Table 1.

MRI Findings & Motor Neuron Disease (NMD)

MRI showed anterior cord compression at the level of C5 and C6 in 19 (27.14%) patients and C6 C7 in 23 (32.8%) patients. 28 patients had compression at C8 T1 level (40%). Among 70 cases of CSM, 11 (15.7%) patients were suspected of having both CSM and MND (assessment by Awaji criteria). These patients were then evaluated with NCS/EMG, and 8(11.4%) patients had NCS/EMG findings of MND.

These patients were followed for 6 months and 4 patients were diagnosed with MND according to El Escorial criteria. NCS/EMG was conducted in the patients, which showed decreased F wave (a late response that follows the motor response) frequency, consistent with motor neuron disease (NMD). The incidence of MND in our study was 5.7%.

Diagnosis through NCS/EMG & El-Escorial Criteria

8 (11.4%) patients were diagnosed with ALS (amyotrophic lateral sclerosis) by NCS/EMG criteria and 4(5.7%) patients were diagnosed as definitive ALS+CSM after 6 months of follow-up using El Escorial criteria.

Findings from Nurick Grading

Our patients had Nurick grade III in 32(45.7%) and Nurick grade IV in 17(24.2%), Nurick grade V in 10(14.2%) patients, Nurick grades I&II in 5(7.14%) and 3(4.2%) patients, respectively. In the remaining 3 patients, the grading was inaccessible due to low GCS.

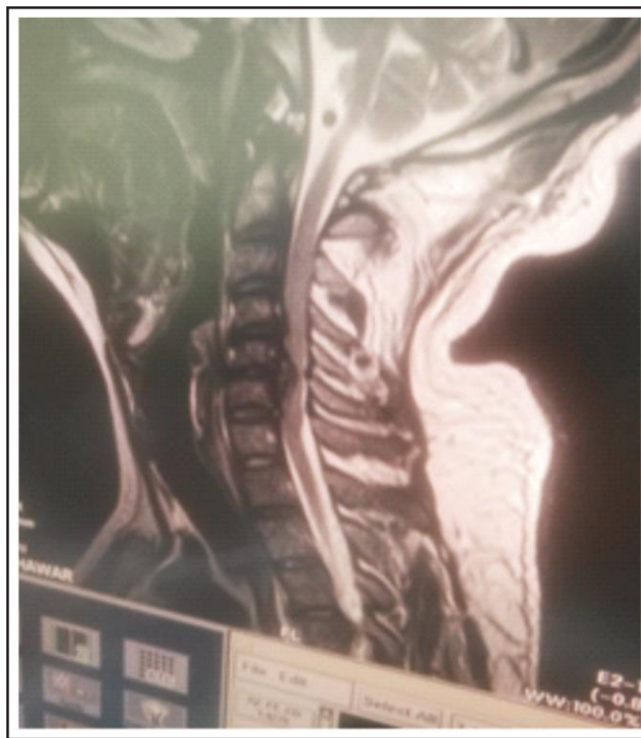


Figure 1: MRI Cervical Spine showing disc prolapse at C5-C6

DISCUSSION

Cervical spondylotic myelopathy and motor neuron disease share common clinical findings and hence they both should be suspected in patients presenting with signs and symptoms of CSM. The majority of our patients (55.7%) had a deltoid muscle grade of 4, whereas 51.4% had a biceps brachii muscle grade of 4. For triceps, the majority of patients (50%) were in grade 3, while for wrist extensors, 45.7% of patients were in

Table 2: Muscle Strength Evaluation in individual muscle groups by MRC scale (from 0-5).

Muscle Group	MRC Grade 0 (Complete Paralysis)	MRC Grade 1 (Minimal contraction)	MRC Grade 2 (Active Movement with Gravity Eliminated)	MRC Grade 3 (Weak Contraction Against Gravity)	MRC Grade 4 (Active Movement Against Gravity & Resistance)	MRC Grade 5 (Normal Strength)
Wrist Extensors	1 (1.4%)	8 (11.4%)	32 (45.7%)	15 (21.4%)	12 (17.14%)	2 (2.8%)
Finger Flexors	10 (14.2%)	26 (37.1%)	20 (28.5%)	11 (15.7%)	2 (2.8%)	1 (1.4%)

Deltoid	0 (0%)	1 (1.4%)	3 (4.2%)	15 (21.4%)	39 (55.7%)	12 (17.1%)
Biceps Brachii	0 (0%)	5 (7.1%)	7 (10%)	10 (14.2%)	36 (51.4%)	12 (17.1%)
Triceps	0 (0%)	3 (4.2%)	10 (14.2%)	35 (50%)	15 (21.4%)	7 (10%)

grade 2. Just 1.4% and 14.2%, respectively, of the wrist extensors and finger flexors were classified as grade 0. Just 17.1% of the deltoid and biceps brachii, 2.8% of wrist extensors, 1.4% of finger flexors, and 10% of triceps were classified as grade 5. In 27.14% of patients, an MRI revealed anterior cord compression at the level of C5 and C6, and in 32.8% of patients, C6 and C7. Compression was present at the C8 T1 level in 28 individuals (40%). Of the 70 CSM cases, 15.7% of the patients had possible dual diagnoses of MND and CSM. El Escorial criterion was used to determine the definite diagnosis of ALS+CSM in 5.7% of patients following a 6-month follow-up, whereas NCS/EMG criteria were used to diagnose 11.4% of patients with ALS. In our study, the incidence of MND was 5.7%. 45.7% of patients had Nurick grade III, 24.2% had grade IV, 14.2% had grade V, while 7.14% and 4.2% of patients had grades I and II, respectively.

In a study by Kleopa et al,²⁴ four patients had bilateral, gradually worsening cramps and fasciculations together with asymmetric weakness and muscular atrophy in the lower limbs. The sensory problems didn't matter. Lumbosacral myotomes, extending from L2 to S1, were determined to be weak. Extensor plantar responses were observed in one patient with proximal leg involvement, and the tendon reflexes varied. The electromyogram revealed segmental chronic and often active denervation limited to the weak myotomes in the lower limbs, although the nerve conduction investigations were normal. All patients' MRIs revealed increased signals in the surrounding cord and signs of spondylotic lumbosacral myelopathy linked to disc herniation at the T11/T12 spinal level. This peculiar solely motor appearance might be the consequence of anterior spinal artery compression-related ischemic myelopathy. All their patients have been sent for potential decompression; in fact, active denervation in three of them's EMGs plainly shows persistent motor neuron damage, which calls for decompression. It could be crucial to

identify this disease early on to reduce the risk of permanent brain damage.

One typical differential diagnosis for spinal onset Amyotrophic Lateral Sclerosis (ALS) is cervical spondylogenic myelopathy (CSM). It may be difficult to diagnose ALS in persons with CSM. According to Torrieri et al, (2021),²⁵ 27.3% of patients had a diagnosis of CSM + ALS and 72.7% of patients had a diagnosis of CSM. Five of the patients (83.3%) who met the criteria for ALS at EMG 18.2% had a clinical follow-up diagnosis of clinically definite ALS + CSM. Comorbidity between CSM and ALS is a condition that occurs often in clinical practice. Because EMG has a high degree of accuracy in identifying ALS, it should be used to better select people who may benefit from surgery in CSM patients. There are certain clinical similarities between amyotrophic lateral sclerosis (ALS) and cervical spondylotic myelopathy (CSM). Therefore, in the differential diagnosis of individuals presenting with CSM signs and symptoms, motor neuron disease (MND) should be considered. Because the symptoms of MND in patients with CSM are distinct, it might be challenging to diagnose the condition solely on overlap. When individuals exhibit contradictory symptoms, more research has to be done. The same patients may experience both MND and CSM. Because MND impacts how well these patients respond to surgery, a workup for MND is therefore necessary for CSM patients. It was shown through a special case study that both diseases coexisted in the same patient.²⁶

Reduced hand dexterity, unstable gait, and sensory and motor dysfunction are among the classic signs of CSM. When a patient has a suspicion of CSM, magnetic resonance imaging is the preferred imaging modality; however, those who are not eligible for it may use computed tomography myelography. While patients with moderate-to-severe illness have surgical treatment, individuals with mild CSM may receive nonoperative care as well. For that reason, it is

advised that any patient suspected of having CSM be sent to a spine surgeon as soon as possible. Delays in diagnosis and treatment might cause permanent impairment.²⁷ The neurodegenerative condition known as motor neuron disease (MND) is chronic, progressive, and now fatal. Even though pain is a common sign of motor neuron disease (MND), it is frequently misinterpreted as another illness when it manifests before weakening sets in. Because of their unusual symptoms, patients are frequently referred to non-neurological departments, which might cause a delay in diagnosis and incorrect therapy. Misdiagnosis can be decreased via careful assessment of the clinical development of the symptoms, thorough EMG and nerve conduction studies, improved clinical approaches to diagnosis, and increased public knowledge²⁸

CONCLUSION

The same patients may experience both MND and CSM. In individuals with cervical spondylotic myelopathy, the incidence of MND is 5.7%. Because MND impacts how well these patients respond to surgery, a workup for MND is therefore necessary for CSM patients. We should perform NCS/EMG in patients with CSM to exclude MND to improve clinical outcomes from surgery.

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Additional Information

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Ethical Review Board Approval: The study was conformed to the ethical review board requirements.

Human Subjects: Consent was obtained by all patients/participants in this study.

Conflicts of Interest:

In compliance with the ICMJE uniform disclosure form, all authors declare the following:

Financial Relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

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AUTHORS CONTRIBUTION

S. No.	Author's Full Name	Intellectual Contribution to Paper in Terms of:
1.	Sahibzada Haseeb Ahmed	Study design and methodology.
2.	Muhammad Kashif Jamal	Literature review and referencing.
3.	Syed Mansoor Shah	Final review and approval.
4.	Bahrul Amin Khan	Data collection and calculations.
5.	Zia Ur Rehman	Interpretation of results.
6.	Syed Mansoor Shah	Analysis of data.