Author's response to reviews

Title: Upper limbs dysmetria caused by cervical spinal cord injury: a case report

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Author's response to reviews: see over
We sincerely appreciated the Reviewer’s comments. The manuscript was revised accordingly in red text and point-to-point responses were also provided.

Reviewer's report:
The case report of LIN et al deals with a 51 year old patient who presented an upper limbs dysmetria after a cervical spine injury.
Case presentation is well described as the mechanism of injury and clinical picture on admission is concerned.
Patient presented a weakness over both upper limbs, with no involvement of the lower, in the absence of sensory deficits, proprioception included. Patient was ataxic while was standing with muscle power normal in his legs. The tendon reflexes of four limbs were increased and no Babinski sign was noticed. The peculiarity of the clinical picture was the presence of bilateral dysmetria of both upper limbs at the finger-nose-finger test, mainly in the left hand.
The Authors emphasize that the dysmetria was out of proportion in relation to the weakness of the upper limbs and the sign was confirmed by three neurologists. Patient was appropriately investigated from the neuroradiological point of view: cervical spine X-ray did not evidence lesions and so was brain CT. Since a brain stem or cerebellar lesion was suspected due to the presence of dysmetria, a brain MR was first performed. No brain stem or cerebellum lesions were evident.
Cervical spine MR was then performed: RM T2-imagings shoved the presence of an hyperintense lesion at the level of C1 segment of the cord. The lesion involved the central area and the lateral column bilaterally. RM imagings also showed a cord compression at the C4-C5 / C5-C6 levels due to disc bulging. At this time the hypothesis of the Authors was that the hyperintense lesion at C1 cord level could have been caused by the trauma and the patient was then submitted to medical treatment (high dose steroids). Steroids resulted in a partial improvement of dysmetria and weakness.
According to the neurosurgical consultation patient was then submitted to C4-C5/ C5-C6 anterior decompression and fusion with cages. Surgical treatment resulted in a dramatic improvement of neurological deficits, including ataxia.

Comments
1) The Authors do not refer whether X-ray of the cervical spine and CT scan of the brain are routinely performed in a patient with a history of cervical trauma
with no obvious clinical sings of brain lesion. The presence of neck pain, weakness of both upper limbs, even in the presence of dysmetria, should indicate that a cervical MR should be performed as a first exam.

Answer:
The presence of neck pain and bilateral upper limbs weakness may be resulted from his cervical spine lesion. Bilateral upper limbs dysmetria was not so common clinically. Considering the possibility of a vascular episode in the cerebellum, which resulted his falling, a brain image was arranged even though there were no other clinical signs of brain lesion at time of examination. (The manuscript was revised from the last 3 lines in page 4 to the first two lines in page 5.)

Comments
2) As for the cord lesion documented at T2-MR C1 cord segment, it is difficult to appreciate in the axial views of Figure 1 the real location of the lesion, whether in the central area and/or at the level of lateral columns or in some other cord areas.
1) As for the cord compression due to disc bulging at the level of C4-C5 / C5-C6, figure 1 does not show with certainty whether the cord was involved and/or the possible presence of hyperdensities within it.

Answer:
We have edited the image. A C1 hyperdensity area was noted to be located in the central area. But there was no obvious hyperdensity area noted at the C4-C6 levels.

Comments
2) The Authors do not report what steroid and the time during which the steroids were administered. After medical treatment, the patient had a clinical improvement, although partial. Since the suggested lesion responsible for the clinical picture was at the level of C1, the Authors do not clearly report what was the indication of surgical treatment of C4-C5 / C5-C6 spine decompression. Besides, the time span between admission and stay in hospital, the beginning and duration of medical treatment, and surgical treatment is not reported.

Answer:
Methylprednisolone therapy was initiated 5 hours after the trauma. The dose given was 100mg/hour (1.6mg/kg/hour) for 48 hours without any additional bolus dose. After the steroids treatment, there was partial improvement but certain portion of the weakness and dysmetria persisted. We then consulted neurosurgeon for surgical intervention and an operation was performed nine days after the trauma. The indication of surgical treatment will be described in the next paragraph.
(The manuscript was revised in line 5-7, page 5 and the last 5 lines in page 5.)

Comments
3) Surgical treatment of C4-C5 / C5-C6 decompression and fusion resulted in a dramatic improvement of symptoms and sings. This is in contrast with the initial hypothesis that C1 cord segment hyperdensity was responsible for the clinical picture, so one can also presume that C1 lesion was not related to trauma. In the case reported by LIN et al a post-operative cervical MR could have probably been able to give some insights on the role of C1 lesion and C4-C5 /C5-C6 cord compression. In clinical practice, patients with cervical spondilosis could present, as a consequence of trauma, MR hyperdensities located within the cord and far from the site of focal spondilotic alterations involving one or more bone and disc segments. Sometimes in these patients, indications to surgical treatment of spondilotic compression at the different level of cord lesion is under question and, usually, patients do not have a dramatic improvement after surgical treatment of spondilotic compression.

Answer:
As your comments, MR hyperdensities in spinal cord may locate within or far from the site of focal spondilotic alternations. Previous report had reported that there was girdle sensation localized from T3 to T11 levels in cases with cervical compressive myelopathy. It was explained by due to ischemia in the watershed zone of spinal cord. Compression of anterior spinal artery at the cervical level might result in ischemic change of watershed zone in spinal cord, even in other spinal levels. (Clinical features of the localized girdle sensation of mid-trunk “false localizing sign” appeared in cervical compressive myelopathy patients, Journal Of Neurology [J Neurol] 2002 May; Vol. 249 (5), pp. 549-53.)

The patient improved after surgical intervention. There were two explanations for his improvement. First, the improvement of patient dysmetria and weakness could be a coincident effect of steroids. The effect of methylprednisolone on motor function could be observed from days to months
Second, the clinical feature of early cord compression is dominant by motor function impairment. It may be resulted from the compression of vessels supplying the spinal cord, causing edema and poor capillary circulation in the watershed zone, which includes the corticospinal pathway. (John Philip Patten. *Metabolic, infective and vascular disorder of the spinal cord.* In Neurological differential diagnosis, 2nd edition. Edited by John P. Patten.) In this patient, the compression of C4-C5 / C5-C6 from the anterior aspect may caused ischemia over the watershed zone of C4-6 level, which involved the corticospinal and rubrospinal tracts. For this reason, we consulted neurosurgeon for surgical management of C4-C5 / C5-C6 spine decompression. His clinical symptoms also improved after the surgical intervention. This may be resulted from relief of spinal artery compression. The surgery also relieved the ischemia change in the watershed zone at the level of focal spondilotic alternations and even at other levels. However, whether the C1 hyperdensity is related to the watershed zone ischemia from C4-C6 compression cannot be confirmed. A post-operative cervical MR could probably give some insights on the role of C1 lesion and C4-C5/C5-C6 cord compression.

(The manuscript was revised from line 5, page 8 to line 10, page 10.)

Discussion of the clinical case deal with an anatomo-physiological review of the brain structures involved in dysmetria. Dysmetria as a sole neurological deficit is well known to be the consequence of a dysfunction of cerebellum and related structures (superior cerebellar peduncles, red nucleus and thalamus). The absence of any involvement of these structures on MR imagines led the Authors to analyze other hypothesis. Since the clinical history was indicative of a spine trauma, the study was aimed to find the role of the spinal cord dysfunction in the presence of dysmetria. As reported by LIN et al the spinocerebellar tract conveys the proprioceptive sensation to the cerebellum, thus influencing coordination and balance.

**Comments**

1) In a patient with spinal trauma, the presence of dysmetria and weakness of the upper limbs associated to ataxia of the lower limbs should discard, as first hypothesis, a cerebellum and/or a brain stem dysfunction.

Two are the spino-cerebellar tracts on both sides of the cord: the anterior that modulate the whole limb position reflexes. In the somatotopic representation of the fibre tracts in the spinal cord, fibres coming from the upper legs are
conveyed laterally. Fasciculus gracilis and cuneatus are also responsible for limbs position. From the clinical point of view, ataxia may be present with dysfunction of these spinal cord tracts. Actually, patient reported by LIN et al had ataxia in the standing position but no sensory disturbances, either deep pressure and proprioception, were evident, although they are usually present with cord ataxia.

**Answer:**
As your comments, the spinocerebellar tract and Fasciculus gracilis and cuneatus are responsible for controlling limbs position. Ataxia may be present with dysfunction of these spinal cord tracts, but dysmetria is not so common. So we proposed the hypothesis that the combination of corticospinal and rubrospinal tract involvement, which lead to the patient bilateral upper limbs weakness and dysmetria.

(The manuscript was revised according to review's comments in line 11-16, page 6.)

The second part of the discussion deals with the role of the rubrospinal tract. The Authors report that the rubrospinal is a descending tract coming from the red nucleus to the rostral part (cervical) of the cord. The red nucleus receive fibres from the interposed nuclei of the cerebellum that – through different connections – influences the corticospinal tract and is involved in ongoing movement of distal extremities.

The conclusion is that rubrospinal tract may play a role in the coordination of limbs. Due to the close location of rubrospinal and corticospinal tract, the co-existence of dysmetria and weakness of the upper limbs may be explained.

**Comments**
1) As reported by the Authors, the role of the rubrospinal tract is not clear in the human beings.

**Answer:**
The role of the rubrospinal tract is not yet so clear in the human beings and the termination level in the cervical spinal cord is also not well known yet.

**Comments**
2) The presence of ataxia of the legs and dysmetria and weakness of the upper limbs in the patient reported by Lin et al could be explained by a postero-lateral dysfunction of the cord. The presence of a post-traumatic lesion
within the central area of the cord is usually responsible for a more severe clinical picture. Furthermore, in these circumstances, a decompression of the spine usually does not permit a dramatic clinical improvement.

3) The presence of dysmetria associated to weakness and ataxia could led to the hypothesis of cortico-spinal, spino-cerebellar and posterior columns involvement. In clinical practice, only a deepen, detailed and sometimes frustrating clinical observation and evaluation could clearly distinguish a true dysmetria co-existing with weakness and ataxia.

**Answer:**

As your comments, the presence of a post-traumatic lesion within the central area of the cord usually will responsible for a more severe clinical picture. (It was revised according to review’s comments in Line 11-12, Page 9) This patient presentation was not so severe, though there was a hyperdensity at C1 level in cervical MR. The improvement after the decompression may result from the relief of spinal artery compression as we have discussed.

The presence of dysmetria associated with weakness and ataxia could lead to the hypothesis of cortico-spinal, spino-cerebellar and posterior columns involvement as your comments. We tried to explain the important role of watershed zone ischemia and rubrospinal tract involvement in this case’s clinical symptoms. More cases are needed to support our hypothesis. (The manuscript was revised in line 15-19, Page 9.)