Case Report

Cruciate paralysis, hypothesis for injury and recovery

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Study Design: Case report and review of the literature.

Objectives: Discuss a case of cruciate paralysis, a review of the literature and the hypotheses regarding the pathogenesis and recovery in spinal cord injuries that cause disproportionate weakness of the upper extremities.

Setting: Thomas Jefferson University Hospital, Philadelphia, PA, USA.

Methods: Case report.

Results: A case of cruciate paralysis is presented involving a 59-year-old female who experienced a gunshot wound to the face. Initial motor exams revealed mild lower limb weakness and absent upper limb function with an upper limb modified American Spinal Injury Association motor score of 0/50 (a modified impairment scale using half point muscle grades). Spinal imaging revealed fractures of the C1 anterior ring and the odontoid process, both associated with multiple bullet fragments. No spinal surgery was performed and she was placed in halo fixation. By 3 weeks she had regained enough upper limb function to manipulate large objects with her left hand and move her right hand. At that time, her upper limb ASIA score was 16/50. By 5 weeks, her upper limb modified ASIA motor score had improved to 31.5/50 and she began manipulating feeding utensils, writing legibly, and brushing her teeth with her left hand.

Conclusions: In this case report we present a patient's motor and functional recovery. We also discuss the hypothesis that the acute central cord syndrome and cruciate paralysis are a likely result of similar pathologic mechanisms and that good functional outcome resulted from an initially disabling trauma.

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Keywords: cruciate paralysis; spinal cord injury; central cord syndrome; tetraparesis; trauma; sci; functional; recovery; outcome

Introduction

Cruciate paralysis (CP) is an uncommon clinical entity which results in a syndrome of disproportionate weakness of the upper compared with the lower extremities. The original case reports by Bell in 1970¹ describe a preferential injury to midline fibers of the upper portion of the pyramidal decussation. This theory maintains that since injury to the cord occurs in a region where upper extremity fibers travel alone after decussation, preferential and reversible injury occurs to the upper limbs. Alternative theories presented obscure the pathologic distinction between central cord injury and cruciate paralysis.² This case report reviews a case of a woman who sustained an

injury to the cervicomedullary junction and developed a syndrome of disproportionate weakness of the upper extremities. Of the 20–30 cases that have been reported over the last several decades only a few detail neurologic progress and fewer still document functional gains. A review of a number of reported cases is shown in Table 1. Detailed description of functional as well as neurologic recovery may lead to greater insight into expected functional outcome and impact our understanding of the pathologic mechanisms of injuries that cause disproportionate weakness of the upper extremities.

Case report

A 59-year-old woman sustained a gunshot wound to her face. Bony injuries included left mandibular fracture, left comminuted fracture of anterolateral ring of C1 and severe fracture of majority of the C2

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body with multiple metal fragments seen surrounding C1 and C2 (Figures 1 and 2). The cord itself was not violated by bone or bullet fragments. Cranial nerve findings included left facial nerve paralysis and left recurrent laryngeal nerve paralysis. The patient was initially moving all extremities, but quickly developed tetraparesis and was intubated for respiratory failure

for 8 days. Her spinal injury was treated non-surgically by halo immobilization. Her upper extremity motor recovery was followed by recording the American Spinal Injury Association motor score³ modified by adding half points for plus grade muscles, ie, 4+ = 4.5 points. This technique has been used previously to identify grades of strength between ASIA motor

Table 1 Cases of cruciate paralysis that have been reported over the last 30 years

Author	Year	Cases	Outcome
Bell	1970	4	■ Two, complete recovery with residual upper deficits ■ One unclear ■ One death from complications
Dimitru and Lang	1986	1	■ Independant ADL's and ambulation with distal upper motor deficits
Marano et al	1986	1	■ Upper spastic paraparesis, normal lower
Erlich et al	1989	1	■ At 5 months complete recovery except clumsiness with hands/fingers
Dickman et al	1990	14	■ Eight complete recovery ■ Five with mild upper extremity only deficits ■ One died of complications
Dai et al	1995	5	■ Three, complete recovery > 6 years ■ One, complete recovery > 2 weeks ■ One, residual upper limb weakness

Figure 1 Lateral radiograph of the cervical spine after gun shot wound to the face. Metal fragments seen at the midline are actually lateral to the cord space

grades.⁴ The patients facial injuries did not allow accurate communication and thus, ASIA sensory examination was not recorded. Initially, her motor score was 0/50 with only mild lower extremity deficits. At 3 weeks, her upper extremity motor score had improved to 16/50. At that time she was able to manipulate large objects and bring a toothbrush to her mouth with adaptive devices with her left hand, but was unable to use her right. Ambulation and transfers were accomplished without assistance. At approximately 5 weeks, her upper extremity motor score was 31.5/50 and she was able to perform functional transfers, could now brush teeth, groom herself and write large letters with her left hand. She still could not manipulate smaller objects such as a pencil or toothbrush without adaptive devices nor was she able

to button a shirt. By 10 weeks, her upper extremity motor score had improved to 40/50 and fine motor skills had improved. At that time she could manipulate smaller objects such as large buttons, hold and write with an adapted pencil and operate a telephone primarily with her left hand. She had difficulty with bilateral hand activities such as opening a screw top bottle and manipulation of smaller objects. Figure 3 illustrates her ASIA impairment score against time and functional landmarks. At 18 months, the patient was again examined. She reported that at 12 months she had regained sufficient coordination to oppose each of her fingers against her thumb slowly and manipulate small objects such as button a shirt with small buttons occasionally using an adaptive device. Examination at 18 months showed essentially normal strength with

Figure 2 Four CT scan images showing the extent of bony injury to the ring of C1 and C2 body. Note that bony fragments are not seen within the cord space

Figure 3 Motor recovery using modified ASIA impairment score *versus* time in days after injury. Upper extremity score is seen in cross-hatched bars and lower extremity score is in shaded bars. Functional landmarks are indicated at the times they were achieved

limited coordination of her fingers with significantly slowed distal dexterity.

Discussion

Cruciate paralysis is a clinical syndrome that results from injury to the spinal cord at the junction with the brainstem. Bell in 1970¹ was among the first to describe this syndrome and propose a mechanism of injury with four case reports. Authors including Marano⁵ and Dumitru and Lang⁶ also reported cases of cruciate paralysis citing the theories of Bell¹ and Wallenberg.⁷ These authors cite the largely unproven theory of somatotopic organization that claims upper extremity fibers cross ventral and rostral to fibers supplying the lower extremities. These fibers are thus more susceptible to mild trauma, such as hyperextension, due to their superficial location. Early descriptions of the acute central cord syndrome by authors such as Schneider^{8,9} suggested that traumas to the cervical spinal cord result in pathologic changes to the central gray matter. An insult which progresses laterally would first injure medially located

arm and hand fibers within the corticospinal tract (CST). This theory depends upon an organized, somatotopic layout of the cervical spinal cord. Recovery of edema in such a case was proposed to occur in a lateral to medial direction manifested by leg, then arm then lastly finger function, which was the pattern he observed clinically. The basis for the somatotopic organization of the cervical cord has been called into question by numerous authors. Studies on monkey spinal cords by Barnard,¹⁰ Nathan,¹¹ Coxe,¹² and Pappas¹³ analyzed the degeneration of selective fiber tracts from cortex or sub-cortex and showed that arm fibers within the corticospinal tract are diffusely intermingled with leg fibers. Bunge¹⁴ and Quencer¹⁵ examined cases of cervical trauma that showed diffuse axonal loss within lateral columns rather than central injury.

Emerging evidence supports a random *vs* a somatotopic organization of motor fibers from brainstem to cervical spinal cord. In addition, cases of mild acute central cord injury occur where the pathologic changes were observed to occur diffusely to the lateral tracts such as the CST rather than a central

injury. It is now suggested that the mechanisms of injury in cruciate paralysis and the acute central cord injury may be similar.² What remains to be explained is the mechanism of injury that results in preferential upper extremity motor deficits.

A review article by Kuypers¹⁶ describes the motor tracts as divided into corticospinal, lateral brainstem and medial brainstem. In multiple studies comparing the impact of lesions of these tracts in the Rhesus monkey, it was proposed that medial tracts represented a system for controlling basic trunk movements and synergistic limb motions. The lateral brain stem system (ie, rubrospinal tracts) allowed for independent gross motion of the limbs. The corticospinal tracts added resolution to the brain stem pathways allowing fine motor control of the distal extremities. Huisman¹⁷ studied the corticospinal tracts (CST) and rubrospinal tracts in the opossum, rat, cat and rhesus monkey found that as one progressed to higher mammals, the rubrospinal tracts became smaller while arm fibers in the CST became larger. This progression suggests an increased dependence upon the CST for fine motor control of the upper extremities as one progresses toward higher mammals. Shinoda,¹⁸ and Phillips¹⁹ both utilized electrical stimulation of peripheral nerves of the baboon forearm to show that a higher proportion of distal motoneurons have monosynaptic connections to the pyramidal tract than proximal motoneurons. Phillips concluded from this work that the structural differences amongst lower and higher mammals lies in the proportion of direct cortical control over distal musculature thereby achieving greater dexterity. Bunge¹⁴ reviewed 22 cases of spinal cord injury and grouped them by pathologic mechanism. Of these 22, four were named 'solid cord' injuries. Three of these injuries displayed no central hematomyelia but showed preferential diffuse axonal disruption in the lateral tracts. Quencer¹⁵ in the same study looked only at central cord injuries and again found the same pattern of injury in milder cord traumas. Only the more severely injured spinal cord displayed central hemorrhage. In these studies, Quencer and Bunge observed that the larger fiber tracts were more dramatically injured than the smaller fiber tracts. Teleologically, it appears that as higher mammals gained greater dexterity by avoiding extensive collateralization, susceptibility to mild, diffuse trauma increased. Recovery of function may be a combination of gradual recovery of larger CST fibers combined with unmasking of lateral brainstem tracts, as proposed by Kuypers and Blight,^{16,20} leading to good gross motor function that precedes recovery of dexterity. In the presented patient, given her pattern of recovery through 18 months, it appears that despite good gross coordination, she never regained many of corticospinal tracts that provide more direct innervation of distal extremity motoneurons. These directly innervated motoneurons likely provide the fast, dexterous movements that she lost even at 18 months.

Conclusion

Injuries causing disproportionate weakness of the upper extremities compared with the lower extremities carry a favorable prognosis for functional recovery. Emerging evidence is blurring the pathologic distinction between cruciate paralysis and the acute central cord syndrome suggesting the need for a single nomenclature for both. Carefully following functional goals in this case allowed for the observation that while gross motor score improved dramatically, dexterity of motor function of the hands appeared to remain impaired even after 10 months. We present a hypothesis for her pattern of recovery. With rehabilitation, patients with these neurologic limitations may still be able to return to a high level of functioning at the time of discharge. We hope that this case report adds understanding to injuries causing disproportionate weakness of the upper extremities.

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