Tohoku J. Exp. Med., 1999, **188**, 95-102

Case Report

A Case of Traumatic High Thoracic Myelopathy Presenting Dissociated Impairment of Rostral Sympathetic Innervations and Isolated Segmental Sweating on Otherwise Anhidrotic Trunk

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SAITO, H., SAKUMA, H. and SENO, K. *A Case of Traumatic High Thoracic Myelopathy Presenting Dissociated Impairment of Rostral Sympathetic Innervations and Isolated Segmental Sweating on Otherwise Anhidrotic Trunk.* Tohoku J. Exp. Med., 1999, **188** (1), 95-102 —— A 3 year-old boy developed flaccid paraplegia, anesthesia below T3 and impaired vesical control immediately after a car accident. Three months later, the pupils and their pharmachological reactions were normal. Thermal sweating was markedly reduced on the right side of the face, neck, and shoulder and on the bilateral upper limbs, and was absent below T3 except for band like faint sweating on T7 sensory dermatome. The left side of the face, neck and shoulder showed compensatory hyperhidrosis. Facial skin temperature was higher on the sweating left side. Cervico-thoracic MRI suggested almost complete transection of the cord at the levels of T2 and T3 segments. We discussed the pathophysiology of the dissociated impairment of rostral sympathetic innervations and isolated segmental sweating on otherwise anhidrotic trunk. ———— high thoracic myelopathy; rostral autonomic innervation; pupil; sweating

Preganglionic neurons innervating the pupillo-dilator are located in (C8), T1 and T2 spinal segments, and those innervating the sweating of the face and neck are in (T1), T2, T3 and T4 segments (Thomas 1926; Foerster 1936; Goetz 1948; Johnson and Spalding 1974).

Cervical or thoracic cord transection usually results in anhidrosis over the cutaneous area innervated by the isolated segments (Thomas 1926; List and Peet 1932; Foerster 1936). Some patients also exhibit dysreflexic hyperhidrosis as a released phenomenon (Head and Riddoch 1918; List and Pimenta 1944; Guttmann 1973). On the other hand, several authors have reported that even

Received October 30, 1998; revision accepted for publication April 18, 1999.

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patients with clinically or morphologically complete transection of the cord do present thermally mediated sweating over the cutaneous area innervated by isolated spinal segments (Seckendorf and Randall 1961; Silver et al. 1991).

Here, we report a child with clinically complete transection of the cord at a high thoracic level, who presented normal pupillary functions, markedly impaired sweating except on the left side of the upper portions of the body, and uni-segmental sweating on otherwise anhidrotic trunk.

Case Report

A 3 year-old previously healthy boy was struck by a car on October 29, 1997. His consciouness remained clear, but the lower extremities became immobile immediately after the accident. He was transferred to a nearby hospital where examinations revealed a transverse fracture of the T3 vertebral bone and right-sided hemothorax. MRI of the cervico-thoracic spine showed no abnormalities of the cord on the day after admission. One week later, however, MRI revealed a high thoracic lesion at the level of the T1 vertebral bone, which was of low signal intensity on T1-weighted images, and its surrounding area was enhanced by Gd-DTPA. On T2-weighted images, the lesion showed high signal intensity, the level of which extended from the C7/T1 disc-space to the middle of the T2 vertebral bone (Fig. 1). On November 13, he was referred to us for rehabilitation.

On admission, he required a wheel-chair and indwelling tube. His mentality, speech, cranial nerves and upper extremities were normal. In particular, the

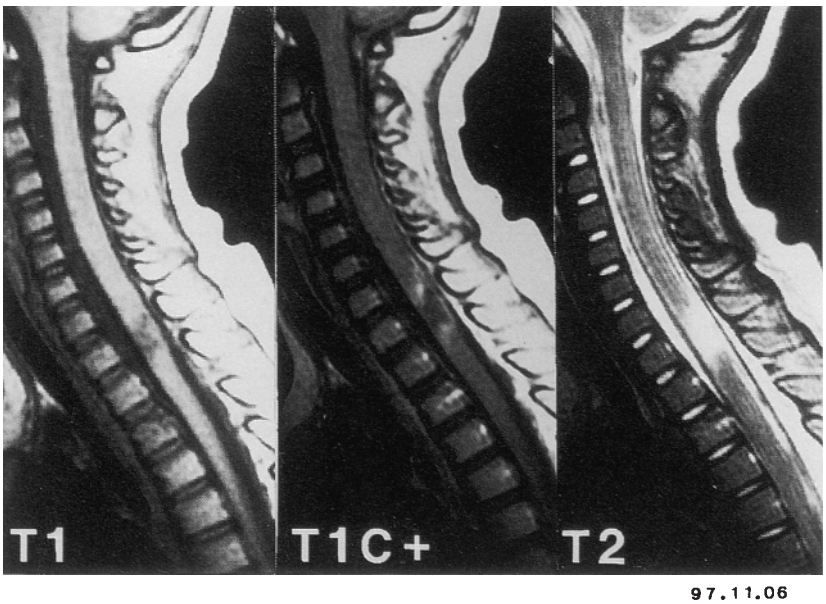

Fig. 1. MRI of the cervico-thoracic portion in the patient 8 days after the accident. T1-weighted images (TR; 500 mseconds, TE; 25 mseconds) show an intramedullary lesion of low signal intensity at T1 vertebral level (left). Its surrounding area is enhanced by Gd-DTPA (middle). In T2-weighted image (TR; 3708 mseconds, TE; 120 mseconds), the cord shows high signal intensity possibly involving T2 and T3 segments (right).

pupils were isocoric with normal reactions and sufficient dilatation in darkness. Lower extremities showed flaccid paralysis with diminished tendon reflex except normal right Achilles tendon reflex. Plantar reflex was extensor and Babinski's sign was positive on the right side. Abdominal skin reflex and cremasteric reflex were abolished. Sensations were lost bilaterally below the T3 level. He showed marked sweating on the left side of the face and neck, especially when he was actively moving a wheel-chair. The trunk and lower limbs were dry, and the feet were warmer than the hands on palpation. The patient had no sensation of bladder filling. The vesical capacity was 50 ml. The volume of active urination was 20 ml, and residual urine 30 ml. Vesico-urethral reflex was not observed. Anal reflex was present on the left side and equivocal on the right side. Bulbocavernous reflex was present on both sides. The patient occasionally cried complaining the right-sided chest pain, which sometimes required analgetic suppository. x-Ray film and CT of the chest failed to detect any abnormalities. The exact nature of the chest pain could not be determined. The lower extremities gradually showed symmetrical spasticity with hyperreflexia, ancle clonus, positive Babinski's sign and triple flexion reflex on pin-prick and thermal stimulation. However, the patient remained totally paraplegic below T3, and required vesical catheterization 5–6 times a day.

MRI on January 12, 1998, showed severe atrophy of the cord at the level of T1 vertebral bone. Both T1- and T2-weighted images showed an area of low signal intensity anterior to the cord from C6 to the upper margin of the T3 vertebral bone. It may have been due to a flow-void in the widened subarachnoidal space (Fig. 2). Repeated MRIs of the lower cervical and upper thoracic segments on May 19 and October 8, 1998 showed same findings, but failed to

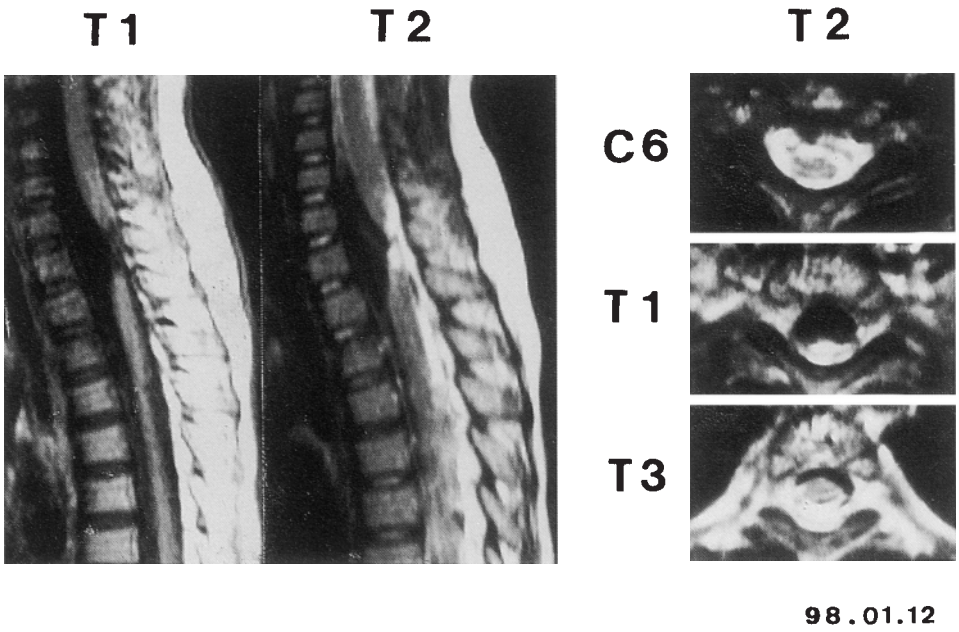

Fig. 2. Three months after the accident, MRI shows marked atrophy of the cord suggesting quasi-complete transection at the level of the T1 verberal bone.

visualize right-left difference in the severity of the structural derrangement.

On March 3, 1999, needle electromyogram showed no denervation activities in gastrocnemius muscles. Motor unit potentials (MUPs) investigated on induced ancle clonus were of normal pattern. No MUPs suggestive of reinnervation were seen. Motor nerve conduction velocity of the left posterior tibial nerve was normal, being 48.2 m per second. Temporal dispersion was not seen. Axon reflex of the cutaneous C-fiber was tested by intracutaneous injection of 0.05 ml of 0.1% histamine phosphate on the lateral surface of the left leg. Fifteen minute after the injection, flare-width tangential to the longitudinal axis of the limb was 54 mm (Control adult subjects without any peripheral nerve lesions: 42.5 ± 5.0 mm, $n = 11$)

Autonomic functions

In supine position, his blood pressure was 110/70 mmHg, and heart rate 72/minute. In sitting position, they were 102/68 mmHg, and 84/minute. Pharmacological pupillary reactions to 5% cocaine and 1.25% epinephrine were investigated using infrared photographs. The pupillary diameters in room-light were 3.5 mm on the right and 3.6 mm on the left, and in semidarkness 6.4 mm on the right and 6.0 mm on the left. Two hours after two drops of cocaine solution, they were 5.5 mm (dilatation ratio [DR]: 1.57) on the right and 4.5 mm (DR: 1.20) on the left in room-light, and in semidarkness 7.5 mm (DR: 1.17) on the right and 7.2 mm (DR: 1.25) on the left. Before epinephrine instillation, the pupillary diameters were 2.5 mm on the right and 2.8 mm on the left in room-light, and in semidarkness 6.3 mm on the right and 6.4 mm on the left. An hour after epinephrine instillation, they were 3.2 mm (DR: 1.28) on the right and 3.5 mm (DR:

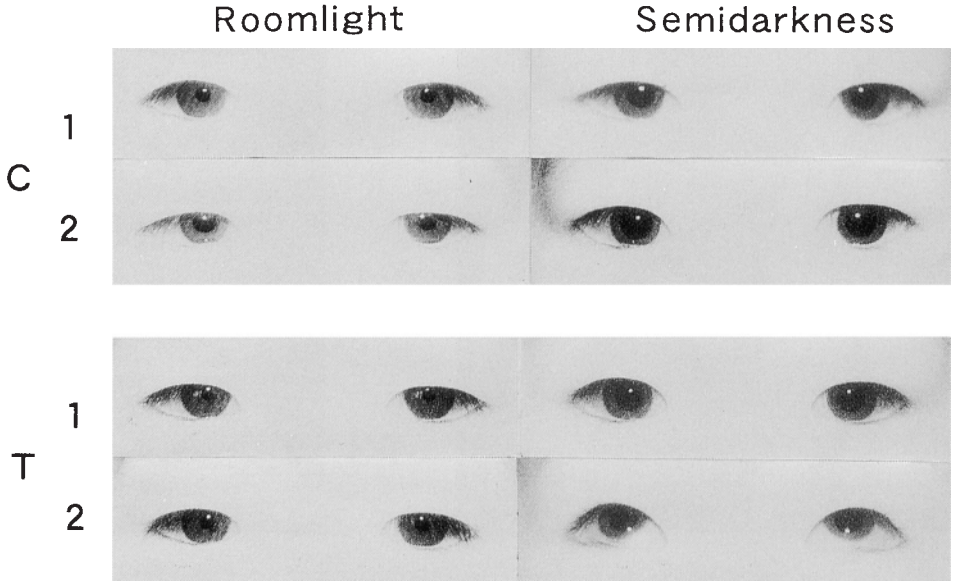

Fig. 3. Pupils are isocoric with normal dilating reactions to 5% cocaine (C) and 5% tyramine (T) (1: before the instillation, 2: two hours after the instillation in C, and 45 minutes after the instillation in T).

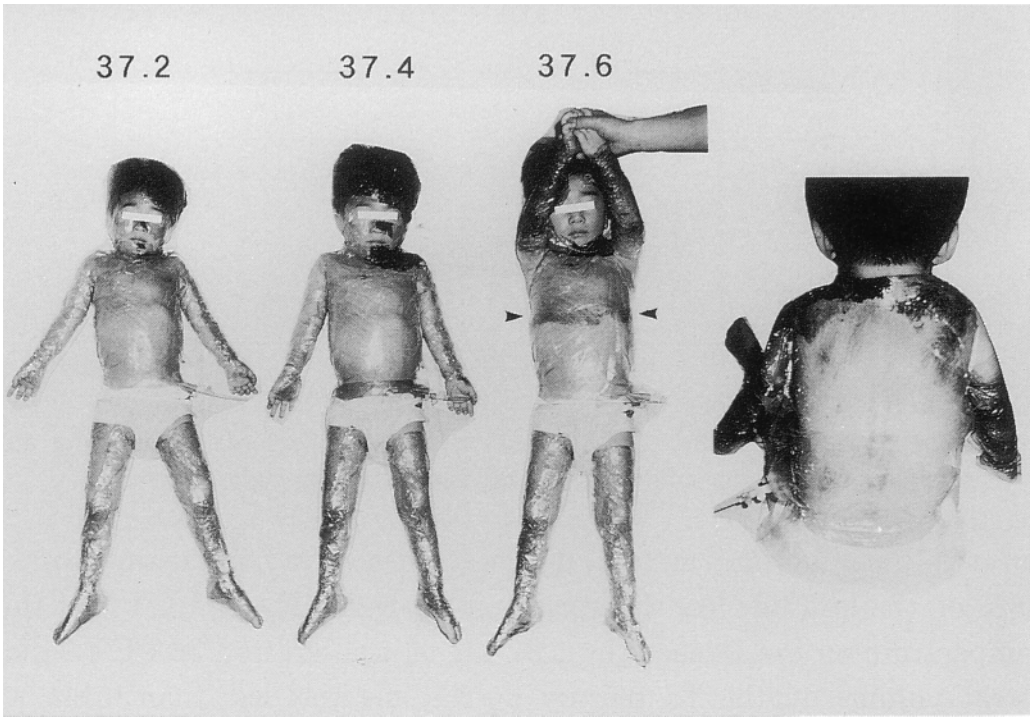

Fig. 4. Thermal sweating is markedly impaired except on the left side of the face, neck and shoulder. Note uni-segmental slight discolaration at T7 level (arrow heads).

1.25) on the left in roomlight, and in samidarkness 6.5 mm on both sides (DR: 1.05 and 1.02) (Fig. 3).

On January 21, 1998, the thermal sudomotor function was examined qualitatively by a modified Minor's colorimetric method (Saito et al. 1990). The patient with a indwelling tube was wrapped with test-sheets in the supine position, and was observed for 30 minutes under a room temperature of 27°C. Deep body temperature was monitored on the forehead by a samister thermometer (Coretemp-CTM-204, probe No: ME-PD5, Termo-Japan Company, Tokyo). Sweating appeared first on the left side of the nose and mental area, then extended to the left side of the face, neck and shoulder. When the body temperature was 37.3°C, heating by electric blankets was started. After heating for 15 minutes, sparse sweating spots appeared on the right side of the face. When the body temperature reached 37.6°C, the left side of the face, neck, and the shoulder showed marked discoloration. The corresponding areas on the opposite side as well as the radial side of both upper limbs showed scattered discoloration. The ulnar side of the arms and the area below T3 were practically anhidrotic except for a slight discoloration limited to the T7 segment. The posterior surface of the body showed similar distribution of discoloration, but the segmental discoloration at T7 was not seen (Fig. 4).

After cessation of heating, sweat volume was measured by a capacitance hygrometer (Hidrograph-AMU-2, Kyokuto-Denshi Company, Nagoya), which revealed apparent sweat expulsions on the left shoulder, but not on the right.

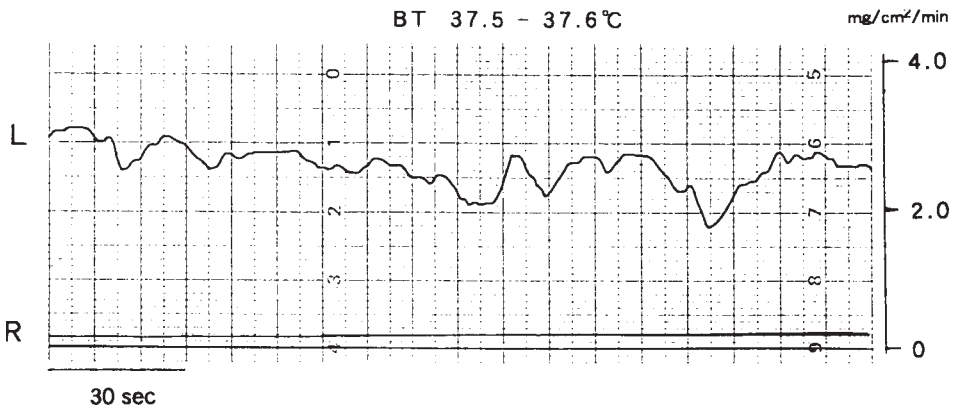

Fig. 5. Hygrometric record of the sweat volume on the forehead showing anhidrosis on the right side. The record is traced from right to left.

The sweat volume (mg/cm^2/minute) on the forehead at a body temperature 37.6°C was 2.820 on the left, but less than 0.282 on the right (Fig. 5). At that time, the skin temperature on the forehead was 37.4°C on the left and 36.4°C on the right. The sweat volume on the T7 sensory dermatome was less than 0.141 without detectable expulsions. On March 3, 1999, the focal sweat volume (mg/cm^2/minute) induced by an intracutaneous injection of 0.05 ml of 5% acetylcholine cloride (ACHS) under room temperature of 26°C was 0.221 on the right side of the T7 dermatomal level and 0.304 on the lateral surface of the right leg (control men of 19–39 years-old with thermally normal sweating: 0.497 ± 0.134, $n = 22$)

Discussion

Reports on the preganglionic innervation of the body surface varies considerably according to investigators, partially because of their different methods of investigations; electrical stimulation, sectioning of the preganglionic fibers, or purely clinical investigation on cord injuries. Still, all authors stressed great inter-individual variations, especially with respect to the most rostral or caudal portions of the body (Normell 1974). The preganglionic neurons innervating the pupillo-dilator are located mainly in the T1 segment, and C8, T3, or even T4 may also contribute to pupillary regulation (Thomas 1926; Foerster 1936; Ray et al. 1943; Goetz 1948; Bonica 1968; Johnson and Spalding 1974). Those innervating the sweating of the face and neck were in the T1-T3 segments according to Foerster (1936), and T2-T4 according to Johnson and Spalding (1974) and Goetz (1948). One common opinion is that the locations of the preganglionic neurons innervating the pupils are one segment higher than those responsible for sweating of the face.

The present patient had flaccid paralysis of the trunk and lower limbs, and lost sensation below T3 and vesical control. Pupillary functions were preserved, but thermal sweating was severely impaired except on the left side of the face and neck, and upper most part of the trunk. MRI showed a high thoracic lesion which involved the T2 segment most severely. Thus, in our patient, the spinal C8

and T1 segments must be largely intact, whereas the T2 segment was preserved on the left, and was damaged on the right. The long ascending and descending tracts relevant to somatic functions, and the hypothalamo-spinal tract which facilitates thermal sweating must have been almost completely transected at the level of T3. The present investigation confirmed previous reports that the relatively high thoracic cord lesions may cause dissociated impairment of the rostral sympathetic innervations (Thomas 1926; Johnson and Spalding 1974).

The uni-segmental sweating on otherwise anhidrotic trunk was an intriguing phenomenon, and may deserve some comments. In patients with relatively high spinal lesions, profuse sweating might be provoked by bladder distension and other viscero-somatic stimuli which enter the cord distal to the lesion (Guttmann 1973). In our patient, however, such a spinal dysreflexia is unlikely because of following reasons. The indwelling tube was used to avoid bladder distension. Muscular spasms and other signs of spinal mass reflex were absent during the test procedure. Moreover, his segmental sweating appeared when the body temperature increased up to 37°C or more, and was no longer observed after the cessation of heating, suggesting that it was thermally mediated one in nature.

It has been generally believed that the transection of the cervico-thoracic cord results in thermal anhidrosis over the area innervated by segments distal to the lesion. However, Seckendorf and Randall (1961) reported that thermally mediated sweating does occur even after a complete cord transection. According to Silver et al. (1991), this type of sweating is distributed sparsely and unevenly without confinement to certain segments, but more apparent on rostral portions than caudal portions. The segmental sweating on the trunk in our patient showed similar characteristics, but was confined to the T7 segment. Since the preganglionic fibers end in multiple paravertebral sympathetic ganglia, the excitation of preganglionic neurons in one segment must provoke sweating of multisegmental distribution (Foerster 1936).

The anhidrosis due to central or peripheral nerve lesions may sometimes accompany hyperhidrosis on neighboring zone, and has been called perilesionary hyperhidrosis or border-zone sweating (Guttmann 1933, 1973; Johnson and Spalding 1974). According to Guttmann (1933), this sweating may occur not only on the side of the lesions but also on the opposite side, and that the area with perilesionary hyperhidrosis are usually painful or hypersensitive to sensory stimuli. Our patient complained pain in the chest, but we could not specify its nature. The patient had hemothorax just after the accident. It is possible that certain intercostal nerve (s) had been damaged, though chest x-ray film failed to reveal a fructure or other lesions which may have caused the irritation of the seventh intercostal nerve.

Thus, at present, we speculate that the uni-segmental sweating on otherwise anhidrotic trunk in our patient may have been the result from the combined effects of high thoracic transection and possible intercostal nerve damage. This

may also explain the absence of the segmental sweating on the back. Further investigations of similar cases are required to elucidate the pathophysiological mechanisms of the autonomic dysfunctions in the spinal cord lesions.

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