CASE REPORT

FIFTY YEARS SURVIVAL AFTER CERVICAL FRACTURE AND FUSION

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Summary. A patient who survived for 50 years after two cervical injuries is described. Among other effects he showed remarkable compensatory changes in the vertebrae above and below a cervical fusion operation. He also developed a meningeal pseudocyst following lumbar laminectomy. The history and pathological findings are considered to be instructive.

Key words: Spinal injury; Pseudarthrosis; Pseudocyst; Survival.

Introduction

LONG TERM survival following cervical spinal cord injury was in the past unusual. It is only in comparatively recent times that a more favourable life expectancy has become commonplace. This improvement is due to many factors. We believe the most important of these to be the multi-disciplinary approach which is practised in specialised units.

The long term survival, the modes of development of the injuries, the late surgical treatment with subjective improvement are some of the unusual aspects of this patient which, supported by the pathological findings, prompted this report. We hope that the record will be of value both clinically and historically to others.

Case Report

Clinical Features

The patient was an American geologist who died in 1980 at the age of 75 years. His initial spinal injury, sustained in 1930, was the result of a fall whilst running, the fall causing his neck to twist sideways. This was followed by numbness and tingling in his hands, trunk and lower limbs and by paralysis of both lower limbs. These symptoms lasted for about 30 minutes. After they settled he continued his work. However, he suffered a persistent right-sided limp.

In 1931, while he was sitting on a camel and trying to remove a thorn from his hand, he fainted and fell off. As he fell he was caught by his partner who observed that the patient's head jerked backwards as the fall was arrested. When the patient regained consciousness some 10 minutes later

he found that he had developed painful tingling in both arms and he also found it was difficult to move both arms and the left leg. Furthermore, the right lower limb was completely paralysed and he was unable to void urine.

His partner, after reading instructions in their first aid kit, (which also contained a rubber catheter), boiled the catheter and using vaseline as a lubricant, drained the bladder. Thereafter the patient managed to void spontaneously but with some difficulty. The weakness of his limbs gradually improved and again he was left with a right-sided limp. Two months later they returned to Perth, Australia where X-rays of his neck were taken but no further treatment was given.

Camels were commonly used for exploration work in the desert areas of Australia in the nineteenth and early twentieth centuries, the terrain being too dry and too rough for horses.

In 1939 he returned to America to do a postgraduate degree in geology at Harvard. During his stay he was admitted to the Massachusetts General Hospital (M.G.H.) for investigation of his walking difficulty as he fell frequently and easily.

The following was extracted from the M.G.H. record (Dr Airey): 'Physical examination revealed awkwardness in his right hand function. He walked with a markedly spastic gait, with scuffing of his right foot. The tendon reflexes in the right arm and leg were more brisk than the left, and there was right patella and right ankle clonus. Both plantar reflexes were extensor. No sensory changes were detected, and no muscle weakness or atrophy noted. X-rays of the cervical spine showed extensive new bone formation above the margins of C5, 6 and 7. The body of C5 was narrowed, and at this point there was a kyphosis.' A diagnosis of either a ruptured disc or spinal arachnoiditis was made, and on 9 January 1939, he underwent cervical laminectomy and fusion from C4-6. The following operation note was made by Dr Jason Mixter (of Mixter and Barr fame):

'On opening the dura it was apparent that the spinal canal was narrowed from front to back at the level of the fourth and upper part of the fifth cervical vertebrae. This narrowing seemed to be related to backward bulging of these vertebral bodies. There was no localised protrusion such as would suggest a fragment of bone or ruptured disc found on either side or in front of the cord. It was deemed inadvisable to attempt to remove any of the backward bulging material, and it was thought that a decompression obtained by leaving out the laminae would be sufficient to relieve pressure on the cord. The dura was closed with silk and then bone flaps were raised from the laminae and osteo-periosteal grafts slid under them on each side.'

Postoperatively the patient considered that he had gained 25 per cent improvement in his walking ability. He later returned to Western Australia to continue his geological work. He married and fathered three children. His subsequent medical history included: lumbar manipulation for sciatica in 1959; myocardial infarction in 1961, prostatectomy in 1962 (which did not significantly improve his difficulty with micturition), and right lumbar sympathectomy in early 1967 for peripheral vascular disease.

In late 1967 he was admitted to the Royal Perth Rehabilitation Hospital for further treatment because of increasing lower limb weakness. Examination revealed incomplete lower motor neurone paresis of both lower

FIG. 1A & B

The X-rays demonstrate the spinal stenosis at the lumbar levels prior to decompressive laminectomy.

limbs but greater on the right, together with weakness and wasting of the intrinsic muscles of the right hand. The bladder paresis now appeared to be also of lower motor neurone type. Myelography revealed multiple level lumbar spinal stenosis, (Fig. 1A & B) so that an L1-4 decompressive laminectomy was performed by Mr J. S. Lekias. At operation the dura was opened and not sutured. The patient's lower limb paresis improved after the operation but over the next 5 years his condition regressed to the pre-operative state. Postoperatively the bladder returned to an 'upper motor neurone type' and his difficulties with micturition persisted. Transurethral resection was performed in 1968 with slight improvement in function. In 1969 ischaemia developed in both lower limbs so that sympathectomy was performed by Mr Robert Paton.

X-rays of his cervical spine taken in 1967 and 1973 showed loss of the normal cervical lordosis, apparent fusion from C2-7, pseudarthrosis between C1-2 posteriorly and degenerative changes at the C7-T1 level (Fig. 2A & B).

FIG. 2A & B

Demonstrates the pseudarthrosis between C1-2 and, by comparing A & B, the movement at this level. Also depicted are the degenerative changes at the C7-T1, the cervical kyphosis, and the posterior spinal fusion from C2-7.

He was next admitted to hospital in 1976 following a supracondylar fracture of his right femur. He subsequently developed gangrene of his right lower limb and an above-knee amputation was performed. Afterward he returned to his home but continued to have problems with micturition and became wheelchair dependent. The last year of his life was spent in the Quadriplegic Centre, R.P.(R.)H. where he died in October 1980 after a further attack of myocardial ischaemia.

His further past history also included malaria in 1928, a haemorrhoidectomy in 1940, and an appendicectomy in 1947.

Necropsy Report

General Findings

A80/202. The body was that of a well nourished Caucasian male weighing 84 kgs and measuring 180 cm in length. The right mid-thigh amputation was noted. Atheroma was present in all major arteries, but was especially severe in the coronary arteries and in the aorta. The right iliofemoral artery was totally occluded by organised thrombus. The heart was enlarged due to moderate left ventricular hypertrophy and weighed 430 grams. The posterior wall of the left ventricle showed extensive recent infarction in

addition to evidence of past healed infarcts represented by large scars.

The lungs weighed 540 gms on the left and 690 gms on the right being heavy and congested. There were also small bilateral pleural effusions which together measured 200 ml. The liver weighed 1870 gms and showed chronic venous congestion with moderate fatty change. The alimentary canal was normal. The kidneys were heavier than normal (L. 150 gm and R. 212 gm) due to a moderate number of senile cysts. The renal parenchyma was normal. The remnants of the prostate were identified together with signs of the previous prostatic operations. There were no other significant findings in the general necropsy.

Neuropathology

The Brain

The arteries at the base of the brain, which weighed 1390 gms, showed advanced atheroma, especially the internal carotid and middle cerebral arteries on both sides. Lacunes due to long-standing ischaemic necrosis were present in the right centrum a semi-ovale, (1.0 cm in diameter) and in the right occipital lobe involving the wall of the calcarine fissure and in which both grey and white matter were lost. This lesion measured 2 cm in diameter. A number of other smaller lacunes each a few mm in diameter were present in the basal ganglia on both sides.

Spinal Column

The normal curvatures of the spinal column were replaced by kyphotic deformity of the cervical spine, loss of the normal dorsal curvature except at T6 where there was a kyphotic angular deformity of about 30°. This dorsal kyphos was due to partial collapse of the T6 vertebral body which was caused by osteoporosis. The normal lumbar lordosis was also lost, the lumbar spine appearing rather straighter than normal.

In the cervical area, there was evidence of the previous laminectomy with deficiency of the spinous processes of C4-5. The laminae in this region were replaced by a thick plate of regenerated bone. This new bone provided the roof to the spinal canal between C4 and 6 and had a more or less 'natural' appearance. When sectioned vertically the discs between C5-6 and C6-7 were found to be obliterated. These lesions corresponded to the early fractures. The loss of the normal lordotic curve of the cervical spine was caused by wedging of the vertebral bodies and fusion operation. However, the spinal canal in the cervical region was not narrowed. The C1-2 pseudarthrosis with articular cartilage, depicted in the X-rays (Fig. 2A & B), was found to lie posteriorly between these vertebrae. There was severe narrowing of the C7-T1 disc space with marked syndesmophyte formation.

The vertebrae of the thoracic and lumbar regions were also sectioned vertically after removal of the roof of the canal. The lumbar vertebral bodies were osteoporotic and the discs were greatly reduced in width especially at L1-2 and L2-3. In addition, there was posterior ridging at the levels of all of the lumbar discs, but this was more severe at L1-2 and L2-3.

FIG. 3A & B

Illustrates the dumb-bell shaped meningeal pseudocyst found in the lumbar region at necropsy and the drops of myodil occupying the cyst during life.

FIG. 4

This demonstrates in cross-section the cyst found in the cauda equina 4 cm below the conus. It is lined by cancellous bone and fibrous tissue and is surrounded by atrophied roots.

H & E $\times 10$.

There was laminal regrowth in the L1-4 region. At the L3-4 level there was a thin plate of bone which contributed to the wall of a cystic cavity. This thin walled cystic space was of dumb-bell shape and measured 2.5 cm in its longest diameter. This cyst was presumed to have arisen as a result of the decompression where the dura was left open and was in fact a meningeal pseudocyst. Figures 3A & B show the lumbar spines and laminae at post-mortem together with the X-ray taken during life depicting the pseudocyst containing myodil.

Spinal Cord

The spinal cord was exposed by opening the dura anteriorly and posteriorly. The anterior and posterior spinal arteries and overlying leptomeninges were intact. In the cervical region there was a constriction at C4 where the spinal cord parenchyma was thinned to about 60 per cent of normal over a length of approx. 2 cm. Transverse section at C4 showed great reduction in size of all 3 columns and the central grey matter was obscured. The C4 roots were atrophied.

The thoracic and lumbar regions of the spinal cord appeared normal to the naked eye. The cauda equina roots were matted together about 4 cm below the conus and in this area there was heavy calcification. On transverse section, the calcified tissue was found to contain a cyst (Fig. 4). The cauda equina roots at this level were considerably atrophied but the higher nerve roots were more or less normal. In cross section the lumbar cord was architecturally normal.

Microscopic examination of the spinal cord showed loss of central grey and adjoining white matter at C4. The lost tissue was replaced by two glial lined cysts one on each side of the central canal. They contained

FIG. 5

The spinal cord at C4 in cross section stained for myelin. It shows the small, astroglial lined, cysts on each side of the central canal which replace the destroyed grey matter and the loss of myelinated tracts from the posterior and lateral columns. Weil's stain $\times 10$.

small numbers of foamy macrophages. There was also great loss of myelinated nerve fibres from both lateral and posterior columns which stained very poorly in myelin preparations (Fig. 5).

At all other levels the posterior columns showed loss of both myelin sheaths and axis cylinders and isomorphic gliosis. This secondary degeneration was caused by the cauda equina lesion.

Discussion

From the patient's description of his initial injury and the later X-ray appearance, the mechanism of the first injury would appear to have been a combination of flexion rotation and vertical compression. His second injury following minimal trauma suggests that there was persisting instability at the affected cervical level.

The residual effects of the injuries included a spastic weak right lower limb, difficulty in walking, 'awkwardness in using his right hand' and difficulty in voiding. These indicated an incomplete tetraplegia with some features of a Brown-Sequard syndrome, following the injuries.

It is interesting that the posterior cervical fusion at operation was described to be from C4 to C6 but radiologically the fusion extended from C2-7. This suggests that when the fusion was performed, the surgical exposure resulted in a more extensive fusion than was intended. Presumably this was due to the stripping of muscles and periosteum from the relevant cervical laminae.

This cervical fusion from C2-7 caused compensatory changes at C1-2 in the form of a pseudarthrosis and further degenerative changes at the C7-T1 level. These changes were related to the loss of movement from C2-7.

This man's multilevel lumbar spinal stenosis was probably caused by segmental instability in the lumbar region. The pathological findings of disc degeneration and posterior ridging in the lumbar region resulted in secondary degenerative changes in the posterior facet joints. These changes both anteriorly and posterolaterally in relation to the dural sac, produced the stenosis.

The increased incidence of meningeal pseudocyst formation following laminectomy when the dura is left open, as happened in this case, is well known; thus Miller and Elder (1968) reported 10 patients who developed meningeal pseudocysts following laminectomy. They noted that in all patients the pseudocyst developed after a tear, cut or perforation of the dura and arachnoid. They also advised that for any patient who developed pain of undetermined origin after laminectomy, myelography should be performed to exclude meningeal cyst formation. Nash *et al.* (1971), who agreed with Miller and Elder's suggested aetiology, described four patients of their own with post-surgical meningeal pseudocysts of the lumbar spine and compared these with 25 other cases in the literature.

In this patient there was good correlation between the necropsy findings and the clinical neurological deficits. The long tract signs were due to the cervical trauma and the posterior column degeneration arose from the cauda equina lesion.

Summary

An unusual case of incomplete tetraplegia following injuries to the cervical spine sustained in 1930 and 1931 is described. The patient died in 1980 from ischaemic heart disease 50 years after the first accident. He was a geologist whose first spinal injury was sustained when he fell while running, and the second when he fainted while sitting on a camel. Subsequently, cervical laminectomy and fusion operations were performed in Boston in 1939 and lumbar laminectomy for spinal stenosis in Perth in 1967. The noteworthy pathological findings were, cervical kyphos and fusion from C2 to 7 with pseudarthrosis between atlas and axis and a meningeal pseudocyst in the lumbar region.

RÉSUMÉ

Un malade qui survécu pendant 50 ans après deux blessures cervicales est discrédité. Parmi d'autres effets, il a montré des changements compensatoires remarquables dans la vertèbre située au-dessus et au-dessous d'une opération de fusion cervicale. Il a développé aussi un pseudokyste des méninges après une laminectomie lombaire. On considère l'histoire et les découvertes pathologiques instructives.

ZUSAMMENFASSUNG

Ein Patient, der fünfzig Jahre nach 2 Nacken Verletzungen überlebte, ist beschrieben. Er zeigte, zwischen anderen Effekten, besondere kompensierende Änderungen in dem Rückenwirbel über und unter der Nacken Fusion Operation. Er hat sich auch eine Membran Pseudozyste, folgend einer Lendenwirbel Laminektomie, zugezogen. Die Entwicklung und die pathologischen Ergebnisse sind als lehrreich zu betrachten.

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