H. Pascal-Moussellard
P. Cabre
D. Smadja
S. Kaidomar
Y. Catonne

Myelopathy due to calcification of the cervical ligamenta flava: a report of two cases in West Indian patients

Received: 31 July 1998
Revised: 10 December 1998
Accepted: 11 January 1999

H. Pascal-Moussellard · Y. Catonne
Departments of Orthopedic Surgery,
University Hospital of Fort de France,
Martinique (French West Indies), France

P. Cabre · D. Smadja
Department of Neurology,
University Hospital of Fort de France
Martinique (French West Indies), France

S. Kaidomar
Department of Neurosurgery
University Hospital of Fort de France,
Martinique (French West Indies), France

H. Pascal-Moussellard (✉)
Service de Chirurgie Orthopédique,
Hôpital Pierre Zobda-Quitman,
97 261 Fort de France Cedex,
Martinique (FWI), France

Abstract Two cases of cervical myelopathy due to calcification of the ligamenta flava (CLF) are described for the first time in black patients from the French West Indies. A pre-operative CT scan differentiated the diagnosis from one of ossification of the ligamenta flava. Micro-analysis on the operatively excised specimen in one patient revealed a mixture of calcium pyrophosphate

dihydrate crystals and hydroxypatite crystals. Poor outcome in one patient contrasting with excellent recovery in the other one, who had undergone posterior decompressive laminectomy, emphasizes the importance of surgery in the management of CLF.

Key words Calcification · Ligamenta flava · Cervical myelopathy · West Indians

Introduction

Calcification of the ligamenta flava (CLF) of the cervical spine causing myeloradiculopathy is a rare disorder occasionally reported in Japanese people [6]. CLF seems to be exceptional in non-Asian populations. We encountered for the first time two such cases in black patients living in the French West Indies. We present here the clinical and radiological findings and outcome in these patients.

Case reports

Case 1

A 72-year-old black woman first noticed numbness in her hands 3 months before admission. During the next weeks, she had diffi-

culty walking and working with her hands. She had no history of spinal injury or rheumatic disease. On admission, the gait was clearly ataxic and neurological exam showed a proximal weakness in the lower limbs. There was no impairment of touch sensation, but position sense, in particular, was absent in all limbs. Deep tendon reflexes were brisk, a bilateral Hoffman sign was found and plantar responses were extensor. She complained of neither radiating pain nor urinary symptoms. General examination was otherwise normal. Routine blood tests were unremarkable. Myelography revealed a partial block at the level C4–C5 and a subsequent CT scan showed voluminous oval masses involving both ligamenta flava at the same level (Fig. 1). The patient was then treated with posterior decompressive C4 and C5 laminectomies. At surgery, the above masses appeared as white granular deposits within degenerated ligamenta flava and were easily dissected from the dura matter. Microscopic examination of the removed material revealed hydroxyapatite crystals (HAP) at the centre of the nodules, surrounded by calcium pyrophosphate dihydrate crystals (CPPD). After surgery, there was dramatic improvement. The gait returned to nearly normal and disturbances of the deep sensation

Fig. 1 Case 1. CT scan at the C4 vertebral level showing two voluminous oval masses involving both ligamenta flava

completely disappeared. At 1-year follow-up, the patient's symptoms had disappeared completely and she was without complaints.

Case 2

A 64-year-old black woman was admitted in February 1996 with complaints of dysesthesia in the distal parts of the arms and gait disturbance. She had an 8-year history of left S1 radiculopathy related to a narrow lumbar spinal canal, which had partially resolved following laminectomy in 1990. About 3 months before admission, after a fall, she noticed dysesthesia in both hands, in the right foot and in the left leg. She then experienced difficulty in sewing and walking. These symptoms gradually progressed until she was unable to walk without two crutches. Neurological exam raised suspicions of an asymmetric quadriparesis below the C5 segment. Pinprick and temperature sensation was impaired in the left lower limb. Deep tendon reflexes were absent at the C5–C6 level, but bilateral triceps brachii reflexes were increased and a bilateral Babinski sign was found. A metrizamide-enhanced CT myelogram strongly suggested asymmetric calcifications of the ligamenta

Fig. 2 Case 2. CT scan following myelography showing compression of spinal cord by a right calcified lesion projecting from the posterolateral aspect into a narrow cervical spinal canal

flava at the level of C5–C6. Compression of spinal cord was consistently observed due to the right calcified lesion projecting from the posterolateral aspect into a narrow spinal canal (Fig. 2). The location of the calcifications was further confirmed by MRI, which also showed an increased signal intensity in the spinal cord on T2-weighted images. Although her neurological status worsened, the patient was reluctant to undergo surgery. During the next months, weakness gradually increased, and impairment of sphincter function appeared in July 1996. At that time, the patient was confined to a wheelchair. She was dependant in all self-care activities and neurological exam showed a left Hoffman sign, giving additional evidence of motor function deterioration.

Discussion

Since the first description by Polgar in 1929 [15], about 100 cases of cervical myelopathy due to CLF have appeared in the literature. Most of them (over 90%) were diagnosed in Japanese patients [1]. Our report of two West Indian patients, as well as a previous one involving a black American [3] suggests the disease may be overlooked in people of African ancestry. The prominent neurological picture is a chronic cervical myelopathy affecting old women with symptoms and signs of impairment of the posterior spinal tracts [1, 6]. CT examination is the key procedure in making a diagnosis of CLF. The calcification usually appears as an oval-shaped lesion located in the paramedian area of the posterior part of the spinal canal [11]. As in our cases, the mid-cervical spine is most frequently involved, especially at C4–C5 and C5–C6 levels [1]. Although ossification of ligamenta flava (OLF) rarely occurs in the cervical spine [8], diagnostic confusion may arise between OLF and CLF. In addition, we recently reported a case of OLF in a black Caribbean patient [14]. However, OLF merely extends medially from the capsulae of the articular process [10] and exhibits a continuity with the bony laminae [13]. Thus, the CT scans in our cases, which showed oval masses partly contacting the lamina and not the characteristic OLF high-density, V-shaped linear structure [14], implied CLF and not OLF.

Cristallographic analysis of CLF identified CPDD, HAP or both [2]. Kawano et al. found that HAP was surrounded by CPPD [7] and presumed the latter occurred first, followed by a transformation of CPDD to HAP, a more stable crystal, in the central part of the flavam. Our findings in case 1 reinforce this hypothesis. However, recent reports favour a wider spectrum of histopathological features, such as HAP in the dural side of the ligamenta flava [4] or acute inflammation accompanying the crystalline deposits [5]. It is commonly accepted that mineralized deposits precipitate around the degenerated elastic fibers of the ligamentum flavum [9] due to a conjunction of various factors including aging process, decrease in oestrogen in older women, mechanical stress of the mid-cervical spine [13] and chondrocytic metaplasia [12].

The peroperative presentation of CLF is a chalky white paste developed inside the degenerated ligamentum

flavum [7], which is always easily dissected from the dura matter. Because most patients described in the literature underwent laminectomy or laminoplasty, little attention has been paid to the natural history of CLF. Lack of surgery in case 2 led to severe tetraparesis within a few

months. On the other hand, the excellent postoperative results in case 1 demonstrate the importance of surgical treatment, which must be conducted before patients suffer irreversible spinal cord damage.

References

1. Baba H, Maezawa Y, Kawahara N, Tomita K, Furusawa N, Imura S (1993) Calcium crystal deposition in the ligamentum flavum of the cervical spine. *Spine* 18:2174–2181
2. Berghausen E, Balogh K, Landis W, Lee D, Wright A (1987) Cervical myelopathy attributable to pseudogout. Case report with radiologic, histologic, and crystallographic observations. *Clin Orthop* 214:217–221
3. Ellman MH, Vazquez T, Ferguson L, Mandel N (1978) Calcium pyrophosphate deposition in ligamentum flavum (letter). *Arthritis Rheum* 21:611–613
4. Hijioka A, Suzuki K, Nakamura T, Yokoyama M, Kanazawa Y (1994) Light and electron microscopy of hydroxyapatite depositions in the ligamentum flavum. *Spine* 19:2626–2631
5. Imai S, Hukuda S (1994) Cervical radiculomyelopathy due to deposition of calcium pyrophosphate dihydrate crystals in the ligamentum flavum: historical and histological evaluation of attendant inflammation. *J Spinal Disord* 7:513–517
6. Iwasaki Y, Akino M, Abe H, Tsuru M, Tashiro K, Miyasaka K, Kaneda K, Isu T, Ito T (1983) Calcification of the ligamentum flavum of the cervical spine. Report of four cases. *J Neurosurg* 59:531–534
7. Kawano N, Matsuno T, Miyazawa S, Iida H, Yada K, Kobayashi N, Iwasaki Y (1988) Calcium pyrophosphate dihydrate crystal deposition disease in the cervical ligamentum flavum. *J Neurosurg* 68:613–620
8. Kubota M, Baba I, Sumida T (1981) Myelopathy due to ossification of the ligamentum flavum of the cervical spine. A report of two cases. *Spine* 6:553–559
9. Kubota T, Kawano H, Yamashita T, Ikeda K, Hayashi M, Yamamoto S (1987) Ultrastructural study of calcification process in the ligamentum flavum of the cervical spine. *Spine* 12:317–323
10. Miyazaka K, Kaneda K, Sato S, Iwasaki Y, Abe S, Takei H, Tsuru M, Tashiro K, Abe H, Fujioka Y (1983) Myelopathy due to ossification of the ligamentum flavum: radiological and histological evaluations. *AJNR* 4:629–632
11. Nagashima C, Takahama M, Shibata T, Nakamura H, Okada K, Morita H, Kubo H (1984) Calcium pyrophosphate dihydrate deposits in the cervical ligamenta flava causing myeloradiculopathy. *J Neurosurg* 60:69–80
12. Ogata M, Ishikawa K, Ohira T (1984) Cervical myelopathy in pseudogout. Case report. *J Bone Joint Surg [Am]* 66:1301–1302
13. Okada G, Hosoi S, Kato K, Ohta K, Tachi Y, Sonoda J, Yada H (1993) Case report 779. *Skeletal Radiol* 22:211–213
14. Pascal-Moussellard H, Smadja D, Cabre P, Raynaud M, Catonne Y (1998) Ossification of the ligamentum flavum with severe myelopathy in a black patient. A case report. *Spine* 14:1607–1608
15. Polgar F (1929) Über interaktuelle Wirbelkalkung. *Fortschr Geb Röntgen* 40:292–298