embolism to the anterior spinal artery, hyperextension injury to the cord by prolonged stupor or coma, and direct toxic effects of drugs.

In our patient spinal fluid pleocytosis was unequivocal evidence of an acute inflammatory reaction in the spinal column. Infection seemed unlikely since cultures showed no growth and other evidences of sepsis were lacking. In view of the immediate sequel and the proximity of the affected segments to the site of injection, a possible entry of the needle into the spinal canal resulting in trauma or inflammation of the cord, due to direct toxic effects of the injected contents were considered. More importantly, neurologically findings were suggestive of an anterior spinal artery syndrome especially as the posterior column sensations were spared. Ischaemia of this arterial territory could have resulted from vasospasm or particulate embolism. Hypotension was not a feature in our patient. Penetration of the needle into the ascending thyracervical or deep cervical arteries in the neck, would have the same effect since these vessels supply radicular branches to the anterior spinal artery. Either trauma or the direct irritant effect of the contents could also induce vasospasm. In addition, methylphenidate (Ritalin) could cause vasospasm by its sympathomimetic action and also, is known to cause necrotising vasculitis. Such adverse effects are not documented with pentazocine (Talwin). Prolonged stupor or coma was also lacking in our patient. The possibility of hypersensitivity and immunological reaction producing myelopathy could not be excluded completely.

The exact pathophysiology of the myelopathy in patients with intravenous drug abuse is not well understood. We feel that the spinal cord ischaemia produced by vasospasm secondary to Ritalin was responsible for the cervical myelopathy seen in our patient. It is important to ascertain the cause of myelopathy in this group of patients so that appropriate therapy is promptly instituted.

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Solitary neurofibroma of the lumbosacral plexus

Sir: There are few reports on the subject of benign neurofibromas in the peripheral nervous system. These tumours have occasion-ally been encountered in patients with multiple neurofibromatosis. Solitary neurofibromas with no other signs at all of von Recklinghausen’s disease, however, are uncommon, rarely painful and most frequently found in the upper limbs. We report such a tumour located in the lumbosacral plexus.

The patient was a 50-year-old woman, whose first experience of pain occurred during a routine gynaecological examination two months prior to hospitalisation in February, 1983. The digital vaginal examination caused a pain that radiated into the back and the right lower extremity. Since the patient’s right breast had been amputated four years previously, for a carcinoma with limited risk of recurrence, an exploratory examination was recommended. On admission to the Department of Gynaecology and Obstetrics, she complained of pain in the inner thigh that was unaffected by posture and pressure. Ultrasound examination revealed a 3.5 × 4.5 cm smooth tumour mass firmly attached to the right wall of the pelvis. Intravenous pyelography demonstrated a soft tissue mass in the right medial pelvic cavity causing displacement of the ureter. During a laparotomy, an encapsulated tumour was found attached to the piriform muscle and enclosing the branches of the deep pelvic veins. Excision of the tumour was not thought to be feasible, and only a hysterectomy was performed with the removal of the adnexes.

In the course of the following weeks, the patient experienced increasing pain, again in her right inner thigh, and her right leg distinctly lagged when walking. Surgical removal of the tumour, which had meanwhile been diagnosed as a benign neurofibroma was therefore undertaken.

The tumour’s upper pole extended to the aortic bifurcation and its lower pole to the obturator fossa, where it was firmly attached to the wall of the pelvis. Dissection began at the level of the obturator fossa. After careful mobilisation, the tumour was totally removed. Pathological examination revealed an encapsulated tumour mass measuring 4 × 4 × 3 cm. On sectioning, the cut surface was of a grey-yellowish colour. Microscopically, the tumour’s capsule was fibrous. The tumour itself was very cellular with scattered fibrilar, cell-poor areas. Elongated oval cells with spindle-shaped nuclei were arranged compactly parallel forming sheets, loose whorls and interlacing bundles (fig 1). There were no atypical nuclei, nor was there any appreciable mitotic activity. In parts, the matrix was myxoid and contained spindle-shaped cells with cytoplasmic cell processes. It surrounded both Schwann cells, many of which contained axons, and bundles of collagen. Axons were found traversing a twisted course within the tumour (fig 2).

Two weeks after operation, the patient was free of pain. Her right leg still lagged slightly but only after prolonged standing or walking. An extremely mild paresis of the adductors, abductors and internal rotators of the right lower extremity was observed. When compared with the left, the right adductor reflexes were somewhat weaker. Neither sensory impairment nor muscle atrophy was found. In conformity with the neurological clinical examination, there was electromyographical evidence of denervation in the adductor magnus, glutaeus medius and tensor fasciae latae. This denervation indicated involvement of the obturator and the superior gluteal nerves, which innervate these muscle
groups as branches of the lumbosacral plexus. Examination three months subsequently, showed the patient's recovery to be excellent. Apart from this benign neurofibroma, evidence of further lesions or stigmata suggesting neurofibromatosis (von Recklinghausen's disease) was not found. Family and medical history was also noncontributory.

On the basis of the histological findings, this tumour can be identified as a peripheral nerve sheath tumour. Most authors have been concerned with the problems of the origin of the tumour cells and an appropriate nomenclature and classification. Comprehensive reviews of the cells from which the tumours arise, and their appropriate nomenclature and classification have been presented by Rio-Hortega and Russell and Rubinstein. In principle, two types of tumours are recognised: (1) the neurofibroma, and (2) the neurilemmoma (schwannoma). Both of these originate from the Schwann cell, the neuroectodermal derivative that sheaths peripheral axons. Pathologically, the neurofibroma is characterised by nerve fibres running through it, as opposed to schwannomas which compress the nerve cell laterally. The neurofibroma is usually a component of the complex entity known as von Recklinghausen's disease or multiple neurofibromatosis. However, these neurofibromas are exclusively plexiform. A tumour with an identical histological pattern, is the solitary neurofibroma, a sporadically occurring neoplasm found in patients with no other signs at all of von Recklinghausen's disease. Russell and Rubinstein doubted the truly "solitary" occurrence of the solitary neurofibromas. Some authors state that all neurofibromas are associated with neurofibromatosis. Harkin and Reed believe such a definition to be incorrect as well as unhelpful, because the majority of patients with a solitary neurofibroma of the skin show no evidence of von Recklinghausen's neurofibromatosis. These authors suggest the existence of a forme fruste of multiple neurofibromatosis. However, when a neurofibroma is diagnosed, the clinician should search for other manifestations of generalised neurofibromatosis. Francis and Glazer reported a patient without diagnostic cutaneous lesions, for whom, however, the diagnosis of neurofibromatosis was still made. This was based on pelvic and lower extremity computed tomography, which revealed numerous tumour masses distributed amongst branches of the lumbosacral plexus and along the right sciatic nerve. Afterwards, it was discovered that two of the patient's sisters had café au lait spots. As this case illustrates, neurofibromas may still be present in the deep tissues of the body even if the characteristic cutaneous lesions are missing. The CT finding of multiple relatively low attenuation soft tissue masses distributed along the course of the peripheral nerves, should allow a diagnosis of neurofibromatosis, even in the absence of the usual cutaneous stigmata.

Solitary neurofibromas are usually found in the skin or subcutaneous tissue, although they have been reported in other sites. In 1927, Borchardt was the first to describe a solitary nerve sheath tumour. Since 1962, solitary neurofibromas have become recognised as a distinct entity following Hearnd's report of 46 peripheral tumours, which included 35 superficial and 11 deep lesions. In 1936, Cutler and Gross observed that the most frequent site was the posterior part of the leg. Stout noted that the most frequent sites of these tumours were the flexor surfaces of the extremities. Pack and Ariel described 303 patients with benign solitary nerve sheath tumours unassociated with the classical type of von Recklinghausen's disease. In this study, solitary neurilemmomas (schwannomas) and solitary neurofibromas were considered the same clinical entity, and the terms were used interchangeably. The authors reported that 44.8% of the tumours occurred in the head and neck region and only 32.6% in the extremities. Only one tumour was found in the pelvis, one in the canal of Nuck and two in the retroperitoneal space.

In the region of the lumbosacral plexus and its branches, the occurrence of a solitary neurofibroma is extremely rare. Robertson et al found an intrapelvic sacral plexus solitary neurofibroma in a 29-year-old woman. Neurologically, no abnormality was identified; the mass was found on palpation, which caused pain that radiated into the right lower extremity. Hudson and Rossi described a solitary presacral neurofibroma. The tumour was found to be firmly attached to the anterior...
surface of the upper sacrum and slightly less firmly to the posterior rectal wall. In a discussion on neurogenic tumours in the sacral region, Adson et al. noted only three benign neurofibromas out of 33 cases. Jackman et al. reported 82 retrorectal neurogenic tumours. Five of these were neurofibromas, which could have arisen from any spinal nerve root and were mainly asymptomatic. Chin et al. found a solitary neurofibroma on a branch of the femoral nerve in two cases. An unusual case of pudendal neuralgia due to a solitary neurofibroma of the perineal region was described by Tognetti et al. The lesion did not appear to be encapsulated, but showed lateral mobility when displaced by the finger. No connections with nerve bundles were apparent, and the nodule was removed.

The morphological features in our case led to the diagnosis of a neurofibroma. Since our patient presented no other neurological deficits, and no skin alterations that could be interpreted as a neurofibromatosis, this tumour may be regarded as a solitary neurofibroma. Due to the neurological deficits found on clinical examination, that is the electromyographical evidence of denervation, this tumour may further be diagnosed as a solitary neurofibroma of the lumbosacral plexus.

Nerve sheath tumours, especially those which arise from a large peripheral nerve, are the often remarkable for the absence of associated neurological deficits. The solitary neurofibromas are entirely benign, display a slowly progressive history and are rarely painful. In these cases, however, careful clinical examination usually reveals the presence of a tumour mass. It is, therefore, extremely important that such peripheral nerve tumours, although rare, are considered when a patient complains of pain and paraesthesia, with or without neurological deficits, in the lower extremities. Furthermore, apart from clinical examination, pelvic CT and ultrasound studies should be performed to relieve the patient’s pain as quickly as possible. Tognetti et al. emphasised the long interval that usually elapsed between the onset of pain and institution of adequate treatment.

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