Five cases of SPS associated with myasthenia gravis have been reported. This is the first report of abnormalities on eye movement recordings strongly suggesting myasthenia gravis in SPS before the patient became seropositive for anti-AChR antibodies. Our patient is probably the third patient with SPS and myasthenia with histologically proven thymoma and the second such patient with positive anti-GAD and anti-AChR antibodies. Our report suggests that patients with SPS can develop other autoantibody mediated disorders even after many months and should be followed up over a long period even if they are asymptomatic. In addition, when patients with SPS have eye movement abnormalities or bulbar symptoms, myasthenia gravis should be suspected even if they are negative for anti-AChR antibodies at presentation. Thymoma should be investigated for, as thymectomy may improve both SPS and myasthenia.

S Thomas

Department of Ophthalmology, University Hospitals Leicester, Leicester, UK

P Critchley, M Lawden

Department of Neurology, University Hospitals Leicester

S Farooq

Ophthalmology, University of Leicester, Leicester, UK

A Thomas

Department of Neurology, University Hospitals Leicester

F A Proudlock

Ophthalmology, University of Leicester

C S Constantinescu

Department of Neurology, University of Nottingham, Nottingham, UK

> I Gottlob Ophthalmology, University of Leicester

Correspondence to: Professor I Gottlob, University of Leicester, RKCSB, PO Box 65, Leicester, LE2 7LX;

ig15@leicester.ac.uk

doi: 10.1136/jnnp.2004.036558

Competing interests: none declared

References

- Vincent A, Grimaldi LM, Martino G, et al. Antibodies to I 125I-glutamic acid decarboxylase in patients with stiff man syndrome. J Neurol Neurosurg Psychiatry 1997;62:395–7.
- 2 Piccolo G, Martino G, Moglia A, et al. Autoimmune myasthenia gravis with thymoma following the spontaneous remission of stiff-man syndrome. Ital J Neurol Sci 1990;11:177–80.
- 3 Nicholas AP, Chatterjee A, Arnold MM, et al. Stiff-person's syndrome associated with thymoma and subsequent myasthenia gravis. *Muscle Nerve* 1997;20:493–8.
- 4 Hagiwara H, Enomoto-Nakatani S, Sakai K, et al. Stiff-person syndrome associated with invasive thymoma: a case report. J Neurol Sci 2001;193:59–62.

5 Saravanan PK, Paul J, Sayeed ZA. Stiff person syndrome and myasthenia gravis. *Neurol India* 2002;50:98–100.

Internal jugular vein thrombosis associated with shiatsu massage of the neck

Thrombosis of the internal jugular vein is a relatively rare condition that can be induced by a variety of mechanical injuries.^{1 2} Acupressure, or "shiatsu", is an oriental massage technique and many acupoints on the body surface, known as "tsubos", are used for shiatsu. Shiatsu of tsubos in the nape of the neck is known to improve tension headache due to neck and shoulder aches. However, we recently came across a case of internal jugular vein (IJV) and cerebral sinus thrombosis after shiatsu massage of the neck.

Case report

A 35 year old man, a non-smoker, was suffering from a stiff neck. He consulted a shiatsu masseur, who performed shiatsu massage on the right side of his neck and right shoulder for 30 minutes. Immediately after the shiatsu massage, the patient noticed pain and swelling of the right side of the neck, both of which subsided within seven days. Two days after the shiatsu massage, he developed a severe, constant right occipital headache and consulted his attending physician. His cervical radiograph was normal. The patient continued to have severe headache; however, and on the seventh day after the massage, he developed blurred vision. On the twentieth day, he developed weakness and paraesthesia of his right arm and leg, and mild agraphia for kanji characters. When he also developed focal motor seizure, he was admitted to our hospital. He underwent a neurological examination on the twenty third day after the shiatsu massage.

The patient did not have any history of recent trauma, dental procedures, or upper respiratory infection. There was no history of any other relevant medication including homoeopathic or herbal medicines, or pathologic conditions. There was no family history of premature stroke or thrombotic events.

Physical examination was normal and no neck mass was detected. On neurological examination, he showed normal consciousness and orientation. Funduscopic examination revealed bilateral papillocdema without haemorrhage, but the remaining cranial nerves were intact. He had mild muscle weakness and sensory deficit in the right arm and leg. Ataxia was not detected in any of the limbs and trunk. Mild agraphia for kanji characters was observed.

Laboratory analysis showed prothrombin time, partial thromboplastin time, antithrombin III, protein C, and protein S were normal, but values for anticardiolipin antibody IgG and lupus anticoagulant were negative. Plasma homocysteine was within normal limits. Autoantibodies and cryoglobulins were absent. No evidence of any systemic disease was found on investigation.

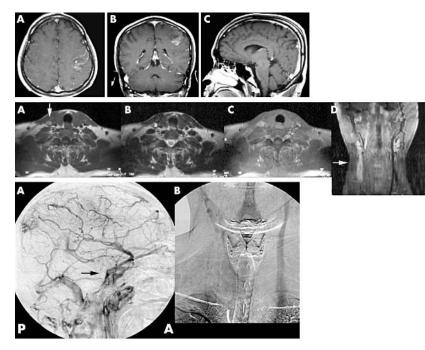


Figure 1 Top panel: post enhancement T1-weighted magnetic resonance (MR) image of the head (A) axial, (B) coronal, and (C) sagittal. (A) and (B) show the left parietal haemorrhagic infarct. The superior sagittal sinus and right transverse sinus show high intensity signal within the lumen instead of the normal "flow void", indicating thrombosis. Middle panel: MR image of the neck (A) T1-weighted, (B) T2-weighted, (C) post enhancement T1-weighted, and (D) coronal T2-weighted showing right internal jugular vein thrombosis without other structural abnormalities (arrows). Bottom panel: digital subtraction angiogram (A) lateral view of the head during the early venous phase of right carotid digital subtraction angiography confirms the non-opacification of the superior sagittal sinus, the deep cerebral venous system and the transverse sinuses. The predominant venous drainage is via the sphenoparietal sinus (arrow). (B) Anteroposterior view of the neck—the right jugular vein had an area of obstruction at its junction with the right subclavian vein.

Cerebrospinal fluid was clear without pleocytosis, but the cerebrospinal fluid pressure was 350 mm H_2O .

Magnetic resonance imaging (MRI) scan of the brain showed infarction with haemorrhage in the left parietal lobe and an area of increased signal intensity in the area of the right transverse and superior sagittal sinuses (fig 1). In addition, MRI of the neck with and without enhancement revealed thrombosis of the right IJV, starting from the junction with the right subclavian vein (see fig 1). However, there were no structural abnormalities adiacent to the right IJV, and the carotid arteries were normal. Digital subtraction venous angiography confirmed extensive thrombosis in the right IJV, the right sigmoid sinus, the right transverse sinus, and the superior sagittal sinus (see fig 1). The rest of the intracranial dural sinuses were patent, and no vascular malformation was detected.

Phenytoin and valproic acid were promptly administered resulting in improvement in the patient's focal motor seizures. He was also given heparin and warfarin and the intracranial hypertension was treated with a lumboperitoneal shunt. The headache and papilloedema slowly improved over the next three weeks, after which the patient was discharged. Neurological examinations over the past several months have revealed only mild clumsiness and paraesthesia of his right hand and leg.

Discussion

Our patient started complaining of a swelling and pain in the right side of the neck immediately after the shiatsu massage of the neck. Subsequently, over a period of about a month, he developed progressive headache, right extremity paralysis, papilloedema, and partial seizures. Although it may be coincidental, the possibility of a causal link between the shiatsu massage and IJV thrombosis is supported by the patient's claim of a massage induced swelling and pain in his neck, and by the temporal relation between the massage and the onset of symptoms that progressed to IJV and cerebral venous sinus thrombosis.

It is difficult to determine the exact mechanism of the IJV thrombosis in our patient. One possibility is that direct trauma or pressure may have induced both venous stasis and vascular injury during the shiatsu massage. The other possibility is that extrinsic compression of the IJV by tissue swelling subsequent to trauma during the shiatsu massage may have induced venous stasis, resulting in thrombosis at this unusual site.

Various forms of trauma have been reported in association with LJV thrombosis, such as jugular thrombosis after catheterisation¹ and Glisson traction for the neck.² To the best of our knowledge, LJV and cerebral venous sinus thrombosis possibly caused by shiatsu massage has not been previously reported. Our case may thus represent a newly identified traumatic aetiology.

Chiropractice is a popular alternative therapy in Western countries, and there are several reports of a relation between chiropractic manipulation and stroke.³ Shiatsu massage, an oriental technique of massage, is a popular alternative therapy in Japan and other Asian countries. Recently this therapy, including the use of a mechanical shiatsutype massager, is becoming increasingly popular in Western countries. In addition, it is generally accepted that this technique is risk free. However, there are two other reports of vascular complications following shiatsu massage in the literature.^{4 5} Tsuboi⁴ reported a case of retinal and cerebral artery embolism directly caused by shiatsu massage of the neck. Elliott and Taylor⁵ also reported two cases of carotid dissection that occurred after use of a shiatsu-type massaging machine. We would therefore like to draw attention to the possibility that shiatsu massage of the neck may cause serious neurological complications.

> Y Wada, C Yanagihara, Y Nishimura Department of Neurology, Nishi-Kobe Medical Center, Hyogo, Japan

Correspondence to: Dr Yuko Wada, Department of Neurology, Nishi-Kobe Medical Center, 5-7-1 Kouji-Dai, Nishi-Ku, Kobe, 651-2273, Japan; wada@nmc-kobe.org

doi: 10.1136/jnnp.2004.038521

Competing interests: none declared

References

- Larkey D, Williams CR, Fanning J, et al. Fatal superior sagittal sinus thrombosis associated with internal jugular vein catheterization. Am J Obstet Gynecol 1993;169:1612–14.
- 2 Simmers TA, Bekkenk MW, Vidakovic-Vukic M. Internal jugular vein thrombosis after cervical traction. J Intern Med 1997;241:333–5.
- Peters M, Bohl J, Thomke F, et al. Dissection of the internal carotid artery after chiropractic manipulation of the neck. *Neurology* 1995;45:2284-6.
 Tsuboi K. Retinal and cerebral artery embolism
- 4 Tsuboi K. Retinal and cerebral artery embolis after "shiatsu" on the neck. Stroke 2001;32:2441.
- 5 Elliott MA, Taylor LP. "Shiatsu" sympathectomy ICA dissection associated with a shiatsu massager. Neurology 2002;58:1302–4.

Congenital dumbbell neuroblastoma mimicking birth trauma

Neuroblastoma is the commonest extracranial malignant tumour in children and neonates.¹ It may involve the vertebral bodies or extend into the spinal canal, compressing the spinal cord, or spread into the retroperitoneal space, involving the lumbosacral plexus. Early diagnosis is important for treatment. We report two cases of congenital neuroblastoma mimicking obstetric related palsies.

Case 1

A 4 month old baby boy with a diagnosis of unilateral leg palsy due to birth trauma, despite normal vaginal delivery, was admitted because of a palpable abdominal mass. The infant's left leg lacked spontaneous movement, was flaccid, and deep tendon reflexes were absent. He had poor rectal tone and dribbling of urine. The levels of urinary catecholamine derivatives were increased. Spinal magnetic resonance imaging (MRI) demonstrated a large retroperitoneal mass with thoracolumbar cord involvement. A diagnosis of neuroblastoma was made following biopsy of the abdominal mass. Multiagent chemotherapy proved effective in reducing the size of neuroblastoma. His left leg function returned after several months' chemotherapy. At present, after two years he is free of disease, he can stand and walk with a brace, and his neurogenic bladder is managed with clean intermittent catheterisation.



Figure 1 Case 2: spinal magnetic resonance imaging scan revealed severe cord compression from T12 to L4 and a large intraabdominal retroperitoneal mass.

Case 2

A 10 day old baby boy was seen for evaluation of right lower limb weakness. He had not moved this leg since birth. He was born at full term via a normal vaginal delivery with vertex presentation. He had a hyperextended thigh and decreased tone in the remainder of the leg. Other limbs were normal. Abdominal examination revealed a palpable mass in the right upper quadrant just lateral to the midline. Sonography of the abdomen revealed a unilateral retroperitoneal tumour adjacent to the right kidney with spinal cord involvement. A spinal MRI showed extensive spinal cord compression from T12 to L4 (fig 1). Biopsy of the paravertebral mass revealed neuroblastoma. The neonate was treated with multiagent chemotherapy. However, he developed paresis of the left leg within two weeks of starting chemotherapy. The spinal cord was therefore surgically decompressed through an osteoplastic laminotomy and the extradural mass was fully resected. Although there was partial recovery of left leg function the right limb remained plegic.

Discussion

Birth trauma causing brachial plexus injury is relatively common where obstetric services are limited, but lumbosacral plexopathy after a normal vaginal delivery is extremely rare. Unilateral lower extremity palsy in a neonate must lead the primary care provider to consider other diseases. The combination of neurological deficits and an abdominal mass should alert the physicians to consider neuroblastoma. Early diagnosis can improve outcome,^{2,3} and neuroblastoma diagnosed even in the prenatal period has been reported to have excellent prognosis.⁴