*Neurology Asia 2010; 15(2) : 133 – 135*

# Morvan's syndrome after intra-scrotal injection of lignocaine and denatured spirit for hydrocoele

Sanjay Sharma *MD (Medicine) DM (Neurology)*, Kamala Kant Bhoi *MD (Medicine) DM (Neurology)*, Pranita Sharma *MBBS*, Sheela Anant *MD (Medicine)*, Ajay Parashar *MS (Surgery) MCh (Urosurgery)*

*Department of Neurology, Modern Medical Institute, Lalpur, Raipur (Chhatisgarh), India*

### *Abstract*

Morvan's syndrome is a rare form of neuromyotonia having prominent central symptoms. We present a series of 9 patients who developed Morvan's syndrome after scrotal tap and local instillation of lignocaine and denatured spirit into scrotal sac to treat hydrocoele. The course of the disease was self-limiting. All patients improved within 3 months of onset of symptoms.

## INTRODUCTION

In 1890 Morvan described a syndrome of myokymia, associated with muscle pain, excessive sweating, weight loss, hallucinations, sleep disturbances and behavioural abnormalities. This form of neuromyotonia is seen in association with autonomic, cognitive and sleep disturbance.[1] We present a series of cases with Morvan's syndrome observed after procedure of hydrocele tapping and instillation of chemical into the scrotal sac to prevent recurrence.

## CASE REPORTS

The diagnosis of Morvan's syndrome was based on both clinical and neuromyotonic discharges on electrophysiology. Nine males with ages ranging between 28 to 47 years, underwent hydrocoele tapping. An equal amount of lignocaine with denatured spirit was injected after syringing to prevent recurrence. The commercially available denatured spirit contains menthol – 1 % w/v and anaesthetic ether I.P. 1 % v/v, and denatured ethanol. Clinical features are summarised in Table 1. The symptoms began between 15 to 45 days after the procedure. All developed excruciating pain in lower limbs and gait disturbance. This was followed by continuous rippling movement involving lower limb. In 3 patients the movement spread upward to involve upper chest and upper limbs. These features were accompanied by marked anxious behavior and decreased sleep. Two patients experienced visual hallucinations. Examination showed brisk deep tendon reflexes and flexor plantar responses. Tremors of outstretched hands were prominent. One patient who had previous poliomyelitis of left lower limb showed no worsening of his weakness.

Electrophysiological tests were performed in all patients. Needle electromyography revealed spontaneous neuromyotonic discharges appearing in doublets and triplets. One patient had mild axonal neuropathy in legs. Non-contrast MRI of lumbo-sacral spine was done in 4 patients. There was partial collapse of L5 vertebra noted in one and was normal in others. MRI of brain was done in one patient (Patient 5) and was normal. There was diffuse slowing in electroencephalogram in one patient (Patient 3). Scrotal sonography suggested enlarged testis and minimal peritesticular fluid. Scrotal exploration was performed in one patient for persistent pain. The scrotal layers were oedematous and fibrosed. The testis was enlarged but fibrosed. On cross-section the seminiferous tubles were replaced by slimy material. The histopathology revealed mature lymphocytes, histiocytes and tangible body macrophages. There was no evidence of tuberculosis or neoplasm. No pus cells or neutrophilic infiltration seen. Antibodies against Voltage-Gated Potassium Channel (VGKC) were not screened for. Patients received intravenous methylprednisolone one-gram daily for three consecutive days and carbamazepine, oxcarbazepine, clonazepam, pregabalin or gabapentin for symptomatic relief. The disease was self-limiting. All improved within 3 month of onset.

## DISCUSSIONS

Neuromyotonia are known to be due to exogenous toxins, certain idiopathic or inherited neuropathies,

*Address correspondence to:* Dr. Sanjay Sharma, Department of Neurology, Modern Medical Institute, Lalpur, Raipur (Chhatisgarh), India. Phone no: 077419001, E-mail:drsanjaysharma123@rediffmail.com

**Table 1: Clinical features of patients with Morvan's syndrome**

| Patient | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 |
|---|---|---|---|---|---|---|---|---|---|
| **Age (years)** | 47 | 40 | 35 | 32 | 30 | 43 | 26 | 33 | 44 |
| **Latency of symptoms after scrotal tapping** | 15 days | 25 days | 3 months | 2 months | 2 months | 1 month | 20 days | 2 months | 28 days |
| **Neuromyotonia in lower limbs** | + | + | + | + | + | + | + | + | + |
| **Neuromyotonia Spreading to chest and upper limbs** | – | – | – | – | + | – | + | – | – |
| **Anxiety, insomnia, diaphoresis** | + | + | + | + | + | + | + | + | + |
| **Visual hallucination** | – | – | + | – | – | – | – | + | – |
| **Epididymo-orchitis** | + | + | + | + | + | + | + | + | + |
| **Concomitant illness** | – | – | Partial collapse of L5 vertebrae | Post polio residual paralysis | Axonal neuropathy | – | – | – | – |

genetic mutations affecting potassium channels, paraneoplastic syndromes and after thymectomy.[2] There were also cases reported after timber rattle snake envenomation, herbicide, lead and mercury poisoning, gold and penicillamine therapy for treatment of rheumatoid arthritis.[3] Recently Panagariya *et al* described multiple cases after ingestion of Ayurvedic drugs.[4] An animal model was produced by exposure to the drug 2, Azaridinyl ethanol[5] and injection of neuromyotonic IgG.[6] The present series presents a previously unreported association of Morvan's syndrome with local injection of spirit and lignocaine into the scrotal sac. The possible explanation in our cases is the formation of antibodies against VGKC evoked by local immune mediated inflammation initiated by denatured spirit. However it was not tested for in our cases. Antibodies against VGKC are detected in about 40% of patients of neuromyotonia with radioimmunoassay using $^{125}$-I-dendrotoxin[7] and also reported with Morvan's syndrome. Previously the central nervous system symptoms are thought to be functional due to absence of abnormalities on imaging studies.[8] It is now believed that anti-VGKC antibodies can affect the CNS in Morvan's syndrome and limbic encephalitis.[9,10] The majority of patients respond to plasma exchange, intravenous immunoglobulin and prednisolone.[11-13] In our series, the disease had a self-limiting course, all patients making a complete recovery.

## ACKNOWLEGEMENT

We are thankful to Dr. Rajesh Gupta and Dr. Sandeep Dave of Ramakrishna - CARE Hospital, Raipur, India for their contribution of cases.

## REFERENCES

1. Serratrice G, Azulay JP. What is left of Morvan's fibrillary chorea? *Rev Neurol (Paris)* 1994; 150(4): 257-65.
2. Cottrell DA, Blackmore KJ, Fawcett PRW, *et al*. Sub-acute presentation of Morvan's syndrome after thymectomy. *J Neurol Neurosurg Psychiatry* October 1, 2004; 75(10):1504-5.
3. de Bray JM, Emile J, Basle M, Morer T, Bastard J. Morvan's fibrillary chorea. *Rev Neurol (Paris)* 1979; 135(11): 827-33.
4. Panagariya A, Kumar H, Mathew V, Sharma B. Neuromyotonia: Clinical profile of twenty cases from North West India. *Neurol India* 2006; 54:382-6.
5. Wallis WE, Van Poznak A, Plum F. Generalised muscular stiffness, fasciculations and myokymia of peripheral nerve origin. *Arch Neurol* 1970; 22: 420-9.
6. Shillito P, Molenaar PC, Vincent A, *et al*. Acquired neuromyotonia: Evidence for autoantibodies directed against K+ channels of peripheral nerves. *Ann Neurol* 1995; 38:714-22.
7. Hart IK, Maddison P, Newsom-Davis J, Vincent

A, Mills KR. Phenotypic variants of autoimmune peripheral nerve hyperexcitability. *Brain* 2002; 125:1887-95.
8. Josephs KA, Silber MH, Fealey RD, Nippoldt TB, Auger RG, Vernino S. Neurophysiologic studies in Morvan syndrome. *J Clin Neurophysiol* 2004; 21:440.
9. Liguori R, Vincent A, Clover L, *et al*. Morvan's syndrome: peripheral and central nervous system and cardiac involvement with antibodies due to voltage gated potassium channel. *Brain* 2001: 124: 2417-26.
10. AT Toosy, SE Burbridge, M Pitkanen, *et al*. Functional imaging correlated of frontotemporal dysfunction in Morvan's syndrome. *J Neurol Neurosurg Psychiatry* 2008; 79{6}:734-35.
11. Newsom-Davis J, Mills KR. Immunological associations of acquired neuromyotonia (Isaacs's syndrome). Report of five cases and literature review. *Brain* 1993; 116:453-69.
12. KM. Tan, VA.Lennon, CJ. Klein, BF. Boeve, and SJ. Pittock. Clinical spectrum of voltage – gated potassium channel autommunity. *Neurology* 2008; 70(20):1883-90.
13. Misra UK, Kalita J, Yadav RK, Agarwal A. Neuromyotonia with neuropathy and muscle hypertrophy: Association or cause? *Electromyogr Clin Neurophysiol* 2006; 46:17-20.