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Case Report

CASE REPORT

Bilateral Idiopathic Carpal Tunnel Syndrome in a 4-Year-Old Girl

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ABSTRACT

Carpal tunnel syndrome (CTS), the most common entrapment neuropathy in adulthood, is rare in childhood. The symptoms may differ to those in adults, or may be misinterpreted owing to children's difficulties in expressing themselves. Cases of idiopathic, bilateral CTS under the age of 5 are rare. A 4-year-old girl presented with pain in both hands and difficulty opening them in the morning. Bilateral severe CTS was determined at electroneuromyography (ENMG). Bilateral wrist splints were advised for both hands. Improvement in ENMG was seen at 2 weeks following conservative treatment.

KEYWORDS: Carpal tunnel syndrome, median nerve, childhood, wrist, splint.

INTRODUCTION

Carpal tunnel syndrome (CTS) is the most common entrapment neuropathy, more common in middle-aged women [1]. Symptoms are frequently (59–87%) bilateral [2]. The clinical picture is characterized by sensory and/or motor symptoms in median nerve distribution in the hand including paresthesias worsening at night, pain and weakness. Although CTS is the most common entrapment neuropathy in adulthood, it is rare in childhood, and the clinical presentation often differs from that in adults [3]. We aimed to present a 4-year-old girl diagnosed with bilateral idiopathic severe CTS, which is rarely reported in the literature.

CASE

A 4-year-old girl presented to our clinic with pain and swelling in both hands, more pronounced in the mornings and on the right side. She had been suffering with pain and the need to move her hands and woke up several times in the night. Her parents reported that she was sleeping with both hands clenched, with difficulty in opening her fingers when she woke up. She was keeping her hands immobile for a long time because of pain, and later opened gradually. She was a well-developed girl with normal weight and height. Physical examination was nonrevealing. Her parents denied frequent use of any mechanical device (smart phone, tablet, etc.). They reported no known cases of CTS in the relatives

Table 1. Median nerve motor and sensory conduction studies

Motor (APB)	Latency (ms)		Duration (ms)		Amplitude (mv)		Conduction velocity (m/s)	
	Right	Left	Right	Left	Right	Left	Right	Left
Wrist	8.5	7.8	6.5	5.0	5.9	6.8		
Elbow	11.2	10.5	6.4	5.1	5.6	6.5	49.1	48.8
Axilla	13.0		6.3		5.1		54.5	
Sensory	Latency (ms)		Peak latency (ms)		Amplitude (μv)		Conduction velocity (m/s)	
	Right	Left	Right	Left	Right	Left	Right	Left
Thumb	NR	NR	NR	NR	NR	NR	NR	NR
Index	5.0	4.4	6.7	5.3	4.9	5.3	20.0	25.0
Third digit	NR	NR	NR	NR	NR	NR	NR	NR

APB: abductor pollicis brevis; NR: non response.

and similar symptoms in the family. Physical examination revealed no findings compatible with arthritis, cervical range of motion was painless and neurological examination was normal. Tinnel and Phalen tests were negative, and no atrophy was observed in the thenar region. Complete blood count, kidney and liver function tests, thyroid stimulating hormone, vitamin B12 and vitamin D were within normal limits including C-reactive protein and erythrocyte sedimentation rate. Rheumatoid factor, antinuclear antibody and cyclic citrullinated peptide were negative.

Roentgenograms of both hands and wrist were normal. Bilateral magnetic resonance imaging of the hands and wrists revealed tenosynovitis of the flexor digitorum superficialis and profundus muscles. Electroneuromyography (ENMG) was scheduled with a preliminary diagnosis of bilateral CTS. Motor nerve conduction studies revealed significantly prolonged median motor distal latencies bilaterally with marked slowing across the wrists. Median sensory responses could not be obtained. Needle electromyography revealed fibrillation potentials in the abductor pollisis brevis muscle with decreased recruitment (Table 1). With the clinical and electrodiagnostic impression, bilateral severe CTS was diagnosed. ENMG testing of parents was also done to exclude a familial CTS, normal results were encountered.

Ultrasonographic evaluation revealed swollen median nerves at wrist region with increased cross-sectional areas, and no mass lesion leading to median nerve compression was determined.

Detailed family history was taken because there is generally an underlying genetic disorder in entrapment neuropathies in childhood. Although our patient did not have any clinical findings suggesting a lysosomal storage disease, lysosomal enzyme activities for several diseases that can cause CTS such as mucopolysaccharidosis and Fabry were investigated. No abnormality was detected. Wrist splints during sleep were advised to prevent the clenching in her hands at night and to ensure rest for the flexor tenosynovitis. Significant improvement was observed at second week follow up, and parents reported that she had never again woken at night and was easily able to move her fingers in the mornings. ENMG on the third month revealed bilateral improvement in median motor and sensory studies (Table 2). Clinical recovery and electrophysiological improvement were achieved, and therefore, the patient was advised to use night splints; she is still under follow up with no symptoms.

DISCUSSION

Childhood CTS was first reported by Martin and Masse in 1958 [4]. The number of cases of childhood CTS in the literature is limited and these take the form of single or small case series.

The majority of cases of childhood CTS involve patients with genetic disorders. Lysosomal storage diseases such as mucopolysaccharidosis and mucolipidosis are most commonly involved in the etiology.

Table 2. Third-month median nerve motor and sensory conduction studies

	Latency (ms)		Amplitude (mv)		Conduction velocity (m/s)	
Motor (APB)	Right	Left	Right	Left	Right	Left
Wrist	5.3	3.8	7.2	8.6		
Elbow	8.5	7.2	7.0	7.8	50.0	48.5
Sensory						
Thumb	NR	3.1	NR	40	NR	NR
Index	4.4	3.3	16	31	22.9	31.5
Third digit	NR	3.5	NR	25	NR	NR

Fifty-two cases of CTS with different etiologies were identified in the review by Poilvache *et al*. The most common cause, in 18 of the 52 cases, was mucopolysaccharidosis and mucolipidosis [5]. Nathalie Van Meir *et al*. investigated 163 cases of childhood CTS between 1989 and 2003. They determined mucopolysaccharidosis in 95 patients, mucolipidosis in 22, familial CTS in 11, trauma in 10, sport-related CTS in 6, fibrolipomatous hamartoma in 4 and idiopathic CTS in 4 cases. Other cases, in the form of single case reports, include the Schwartz-Jampel syndrome, melorheostosis, Leri's pleonosteosis, Dejerine-Sottas syndrome, Weil-Marchesani syndrome, intraneural perineuroma, hemangioma of the median nerve, trigger finger, Klippel-Trénaunay syndrome, scleroderma and Poland syndrome [6].

Intensive sportive activities may be a risk factor for idiopathic CTS in children [7]. There have been case reports of CTS developing after intensive basketball training [8] and skiing [9].

Early diagnosis of CTS in childhood especially under the age of 5 is challenging owing to mild or vague symptoms and mostly owing to inability of the child to clearly describe the symptoms. The delay in diagnosis of up to 6 months is owing to the clinical presentation differing from that in adults. In contrast to adults, the benefit of provocation tests in diagnosis is uncertain. Tinnel and Phalen tests were negative also in our case. Diagnosis must be confirmed with ENMG in suspected cases [3, 10].

Steroid injections and night splinting form the mainstay of conservative treatment in the management of CTS. Surgical treatment is generally recommended for those with severe CTS or when conservative treatment fails. However, owing to the variability in the course of CTS, optimum recommended time for conservative treatment has not been clearly determined [11]. In a review including 20 patients (mean age: 14.4 years) with 31 involved wrists, Batdorf *et al*. recommended a stepwise treatment strategy in childhood CTS starting with wrist splint and proceeding to steroid injection if symptoms do not resolve [12].

Clinical response was achieved on the second week of splint use in our patient and improvement in nerve conduction was confirmed with ENMG. The patient continued using the splints with no recurrence of symptoms at 3-month follow-up.

CONCLUSION

Although idiopathic CTS is a rare condition in childhood, it should be kept in the differential diagnosis of wrist pain in children. Conservative treatment with wrist splints should be the initial treatment of choice.

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