Bilateral Brachial Neuritis Following Diphtheria, Tetanus and Pertussis Vaccination: Case Report

Difteri Tetanoz Boğmaca Aşısı Sonrası Bilateral Brakiyal Nörit

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ABSTRACT Brachial plexopathy in children is commonly caused by obstetrical complications. Only few cases were reported as brachial neuritis which is also known as Personage Turner disease. Antecedent infection, viral diseases, trauma, surgery, and immunization may be the possible etiologic factors causing brachial neuritis. In this case report, a 6 months old child who was referred to our outpatient clinic with bilateral hand, arm and shoulder weakness which occurred after immunization with diphtheria, tetanus and pertussis (DTP) vaccine at 5 months of age is presented. The case was diagnosed as bilateral brachial neuritis according to clinical and electrophysiological data and no possible etiologic factors could be associated with the clinical condition except DTP vaccination.

Key Words: Brachial plexus neuritis; immunization; diphtheria-tetanus-pertussis vaccine

ÖZET Çocuklarda brakiyal pleksopati sıklıkla obstetrik komplikasyonlara bağlı gelişir. Personage Turner hastalığı olarak da bilinen brakiyal nörit çocuklarda çok az olguda bildirilmiştir. Önceden geçirilmiş enfeksiyonlar, viral hastalıklar, travma, cerrahi ve aşılama brakiyal nörite neden olan etiyolojik faktörler arasında yer alabilmektedir. Bu olgu sunumunda polikliniğimize beş aylıkken yapılan difteri tetanoz boğmaca (DTB) aşısı sonrası bilateral el, kol ve omuz güçsüzlüğü şikayeti ile getirilen 6 aylık bir çocuk hasta sunulmuştur. Olguya klinik ve elektrofizyolojik bulgulara göre ve DTB aşısı haricinde bu klinik durum ile bağlantılı olabilecek diğer muhtemel etiyolojik faktörlerin dışlanması ile bilateral brakiyal nörit tanısı konulmuştur.

Anahtar Kelimeler: Brakiyal nörit; aşılama; difteri-tetanoz-boğmaca aşısı

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Brachial neuritis, also known as Personage Turner disease, neuralgic amyotrophy, paralytic brachial neuritis, acute shoulder neuritis, acute scapula humeral palsy or brachial plexus neuropathy is a clinical disorder characterized by the acute onset of severe pain in the shoulder and upper arm, followed by the rapid development of arm weakness in 3 to 10 days. The incidence of brachial neuritis has been estimated at about 1.64/100000 in Rochester, Minnesota.[1] Brachial neuritis affects wide spectrum of age groups ranging from 3 months to 84 years.[2] Although different precipitating factors such as infection, trauma, surgery, immunization and autoimmune mechanisms have been suspected, the etiology can not be identified many times. A study that investigates the clinical spectrum of neuralgic amyotrophy in 246 cases from all age groups revealed that 53.2% of the

patients reported an antecedent event; and five of the cases were due to vaccination.[3] Even though immunization has enabled significant success on various diseases, rarely seen serious adverse reactions or side effects lead to concerns in community. In this case, report a child with bilateral brachial neuritis following immunization with diphtheria, tetanus and pertussis (DTP) vaccine is presented and the literature about this rare condition is reviewed.

CASE REPORT

The 6-month-old child was referred to our pediatric rehabilitation outpatient clinic with lack of bilateral arm and hand movement. In the history; the patient was born in term by vaginal delivery without an obstetrical complication. When he was 2 months old, he was hospitalized with symptoms of acute bronchitis and received ampicilin and gentamycin treatment. After this treatment he was discharged with complete recovery. The patient had been in routine immunization program including two doses of hepatitis B vaccine, one dose of BCG, and two doses of DTP. At 5 months of age, second dose of DTP vaccine was performed in a health clinic of government in our city. After a single vaccination which had been applied to the deltoid muscle, he had fever for few days and stopped moving both of his shoulders, arms and hands. There was no history of trauma or a traction injury before the symptoms started. The laboratory tests including renal function tests, liver enzymes, serum electrolyte and glucose levels were normal. Complete blood count detected hypochromic microcytic anemia with low ferritine levels. He was referred to our clinic for the rehabilitation program one month after the symptoms had started. In physical examination, prominent findings were the absence of movements of bilateral upper extremities. There was bilateral complete loss of shoulder motor functions and partial motor loss at the elbow level. The hand movements were slightly decreased in both hands. The deep tendon reflexes were hypoactive in both upper extremities. Decreased responses to painful stimuli were observed in both upper extremities. Chest X-ray and magnetic resonance imaging of cervical spine and brachial plexus were normal. The electromyography (EMG) performed one month after the disease onset was consistent with bilateral brachial plexopathy with severe axonal degeneration of the upper and median trunks and partial axonal degeneration of the lower trunks (Table 1). There were intensive spontaneous activities in the affected muscles, and sensorial nerve conduction studies of lateral and median cutenous nerves could not be obtained. The physical examination and laboratory findings did not reveal any abnormalities for other etiologic factors such as metabolic, infectious or traumatic disorders. With these findings; the case was thought to be bilateral brachial neuritis secondary to DTP immunization. There were no vaccine related problems including transportation or storage procedures or other significant adverse reaction during the same period according to the health clinic of government information. Therefore, we thought that this was an isolated case.

The child underwent outpatient rehabilitation and home based exercise program which consisted of range of motion and neurofacilitation exercises. The patient could move his left hand and elbow but could not achieve shoulder abduction more than 30°. There was no significant improvement in his right upper extremity. The control EMG which was performed at the fifth month of disease onset showed that severe axonal degeneration in upper trunks of both extremities had been continued (Table 2). In the left upper extremity recruitment of median trunk innervated muscles was improved when they were compared to the first EMG findings. In the right upper extremity, severe axonal degenerations of upper and median trunks were detected. There was no improvement in the control EMG for right arm. The electrophysiological parameters were in accordance with clinical findings. The patient was consulted to pediatric orthopedic surgeons and they suggested carrying on the conservative treatment until two years of age. The patient was assessed at the first year of disease onset but the clinical findings were not different from the fifth month assessment. The patient's mother's consent was taken for publishing scientific data of the child.

TABLE 1: Electroneuromyographic findings at the first month.

Nerve conduction studies						
Nerve	**Recording site**	**Onset (ms)**	**Amplitude (mV-μV)**	**Distance (cm)**		**Velocity (m/s)**
R Median sensory	3rd digit-wrist	1.1.5	14.8	6		40
L Median sensory	3rd digit-wrist	1.5	28.8	6		40
R Ulnar sensory	5th digit-wrist	1.4	7.6	5		62.5
L Ulnar sensory	5th digit-wrist	1.65	13.5	6		36.4
R Lateral cutenous sensory		SNAP can not be obtained				
L Lateral cutenous sensory		SNAP can not be obtained				
R Medial cutenous sensory		SNAP can not be obtained				
L Medial cutenous sensory		SNAP can not be obtained				
R Radial motor	EIP fore arm	2.85	1			
	Axilla	5.5	1.6	12.5		47.2
L Radial motor	EIP fore arm	2.5	4.9			
	Axilla	5.4	3.7	14		48.3
L Median motor	APB wrist	1.8	3.7			
	elbow	3.75	6.1	10		51.3
R Median motor	APB wrist	2.15	2			
	elbow	4.2	1.7	9.4		45.9
Electromyography						
	Spontaneous			**MUAP**		
Muscle	**Fib.**	**PSW**	**Amplitude**	**Duration**	**Polyphasy**	**Recruitment**
R Deltoid	3+	3+	No voluntary activity			
R Biceps	3+	3+	No voluntary activity			
R Triceps	3+	3+	No voluntary activity			
R EDC	3+	3+	N	N	N	Single ossilation
R FCR	3+	3+	No voluntary activity			
R APB	3+	3+	N	N	N	Submaximal
R ADM	None	None	N	N	N	Submaximal
L Deltoid	3+	3+	No voluntary activity			
L Biceps	3+	3+	No voluntary activity			
L Triceps	3+	3+	No voluntary activity			
L EDC	None	None	N			Decreased
L FCR	3+	3+	N	N	N	Decreased
L APB	3+	3+	N	N	N	NA
L ADM	+	None	N	N	N	NA

R: right, L:left, SNAP: sensory nerve action potential, EIP: extensor indicis proprius, APB: abductor pollicis brevis, MUAP: motor unit action potential, Fib.: fibrillation, PSW: positive sharp wave, EDC: extensor digitorum communis, FCR: flexor carpi radialis, ADM: abductor digiti minimi, 1DI: first dorsal interosseous

DISCUSSION

Immunization is generally applied to healthy persons for preventing diseases. Diphtheria-pertussis-tetanus vaccine has been used routinely sice 1940's and dramatically decreased the incidence of these diseases among children all over the world.[4] Although immunization has successfully reduced the incidence of vaccine-preventable diseases, vaccination can cause minor and, rarely, serious side effects or adverse reactions. Therefore several countries develop reporting systems to monitor these side effects. The important issue is, when a side effect is considered, it is usually difficult to attribute the clinical condition directly to vaccine or a coincidental disease. The diagnosis usually is based on clinical suspicion and many times there are no supportive data. Usually there are no specific clinical signs, pathologic findings, or laboratory tests which can determine whether the illness is caused by the vaccine, and moreover these side effects are extremely rare and appropriately designed large studies are not available.

Institute of Medicine (IOM), an independent research organization chartered by the National Academy of Sciences, uses a classification to des-

TABLE 2: Electroneuromyography at the fifth month.

Nerve conduction studies						
Nerve	**Recording site**	**Onset (ms)**	**Amplitude (mV-μV)**	**Distance (cm)**	**Velocity (m/s)**	
R Median sensory	3rd digit-wrist	1.65	23.5	6	36.4	
L Median sensory	3rd digit-wrist	1.72	25.7	6	34.9	
R Lateral cutenous sensory		SNAP can not be obtained				
L Lateral cutenous sensory		SNAP can not be obtained				
L Median motor	APB wrist	3.15	1			
	elbow	3.75	1	8.5	60.7	
R Median motor	APB wrist	2.05	2.14			
	elbow	4	2.42	9	47.4	
Electromyography						
	Spontaneous			**MUAP**		
Muscle	**Fib.**	**PSW**	**Amplitude**	**Duration**	**Polyphasy**	**Recruitment**
R Deltoid	2+	2+	No voluntary activity			
R Triceps	2+	2+	No voluntary activity			
R 1DI	None	None	N	N	N	Submaximal
L Deltoid	3+	3+	No voluntary activity			
L Triceps	None	None	↑	↑	↑	Decreased
L FCR	3+	3+	N	N	N	Decreased
R FCR	1+	1+	↑	↑	↑	Decreased

R: right, L:left, SNAP: sensory nerve action potential, EIP: extensor indicis proprius, APB: abductor pollicis brevis, MUAP: motor unit action potential, Fib.: fibrillation, PSW: positive sharp wave, EDC: extensor digitorum communis, FCR: flexor carpi radialis, ADM: abductor digiti minimi, 1DI: first dorsal interosseous.

cribe the relationships between the vaccines and specific adverse events while reviewing scientific and other evidence. In this classification the side effects are assessed in five categories as follows; no evidence available to establish a causal relationship, inadequate evidence to accept or reject a causal relationship, evidence favored rejection of a causal relationship, evidence favored acceptance of a causal relationship, evidence established a causal relationship. In 1994 IOM review examined events after administration of vaccines (i.e., diphtheria and tetanus toxoids and measles, mumps, hepatitis B, Haemophilus influenzae type b [Hib], and poliovirus vaccines). According to this review, various neurological side effects including seizures, demyelinating diseases of the central nervous system, mononeuropathy, encephalopathy, subacute sclerosing panencephalitis, optic neuritis, transverse myelitis, Guillain-Barré syndrome and brachial neuritis can be seen after vaccination with different causal relationships. For DTP, seizures, demyelinating diseases of the central nervous system and mononeuropathy are the reported adverse reactions with inadequate evidence to accept or reject a causal relationship. Encephalopathy has the evidence that favored rejection of a causal relationship, and Guillain-Barré syndrome and brachial neuritis have the evidence that favored acceptance of a causal relationship according to IOM. IOM concluded that there was the evidence of an association between DTP and chronic nervous system dysfunction in children who had had a serious acute neurological illness after vaccination with DTP, and proposed three possible explanations for this association. First, the acute neurological illness and subsequent chronic nervous system dysfunction might have been caused by DTP. Second, DTP might trigger an acute neurological illness and subsequent chronic nervous system dysfunction in children who have underlying brain or metabolic abnormalities. Such children might experience similar chronic dysfunction in the absence of DTP vaccination if other stimuli (e.g., fever or infection) are present. Third, DTP might cause an acute neurological illness in children who have underlying brain or metabolic abnormalities that would inevitably have led to chronic nervous system dysfunction even if the acute neurological illness had not developed.[5]

To our knowledge two case reports are available in the literature on brachial neuritis following DTP vaccination. Martin and Weintraub reported a

case with brachial neuritis and seventh-nerve palsy after DTP immunization in a 5-month-old child, and attributed it to the pertussis component.[6] Hamati-Haddad and Fenichel described two cases after DTP vaccination.[7] In these cases, clinical improvement occurred rapidly and the condition was described as benign because of spontaneous resolution.[7] The prognosis in our case is not good and differentiates him from the other cases in the literature. We believe that initial intensive axonal degeneration demonstrated with electrophysiological studies may be the predictor of the poor outcome.

The treatment in brachial neuritis should be planned with multidisciplinary approach. Physiotherapy is performed to prevent joint contractures due to muscle imbalance, to strengthen recovering muscles, and to achieve developmental milestones in the presence of incomplete neural and muscular function. Microsurgery, surgical reconstruction of joint problems and tendon transfers can be performed as surgical treatment options. In our case, the orthopedics department concluded to wait until two years of age with routine follow up and planned to perform serial tendon transfer procedures thereafter.[8]

The etiology of brachial neuritis can not be determined many times. However the physician should keep in mind that immunization is one of the antecedent factors, and that brachial neuritis following immunization may have a poor prognosis.

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