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Case Report

Celiac disease presenting as peripheral neuropathy- A rare case report

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Abstract

Celiac disease is an autoimmune disorder usually presenting with gastro-intestinal symptoms and very rarely presents as extra gastro-intestinal manifestations like peripheral neuropathy, arthritis and gluten ataxia. This is a case of an 8 year old girl, diagnosed with celiac disease who developed weakness in upper and lower limbs after she resumed gluten containing diet. Nerve conduction studies showed pure motor polyneuropathy involving all limbs with raised anti- tissue transglutaminase levels (more than 10 times of upper normal limit). Child was put on gluten free diet and symptoms improved after that. Child was still on regular follow up and doing well. Celiac disease should be kept in differential diagnosis if any child present with features of peripheral neuropathy.

Keywords: Peripheral neuropathy, Gluten free diet, IgA Transglutaminase levels, Pure motor neuropathy

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1. Introduction

Celiac disease (CD) is a genetically transmitted autoimmune disorder in which gluten acts as an allergen leading to malabsorption because of damage to intestinal mucosa.[1] Its prevalence is 1% in general population.[2] Most common manifestation of celiac disease is the involvement of GIT, but rarely presents as extra-intestinal manifestations like failure to thrive, dermatitis herpetiformis, arthritis, gluten ataxia, aphthous stomatitis and peripheral neuropathy.[3] These manifestations are most commonly seen in adult population with incidence of 7.4%. However in pediatric age group, incidence is only 0.1%.[1] Herein, we report a case of 8 year old child who presenting as peripheral neuropathy as onset features of celiac disease.

2. Case Report

An 8 year old girl child brought to Pediatrics OPD with complaints of weakness in bilateral lower limbs since last 1 month and weakness of bilateral upper limbs since last 15 days. Weakness in lower limb was symmetrical and gradually progressive. Initially foot went ahead of slippers while wearing and history of repeated falls was present. Patient also had difficulty in standing from sitting position (Gower sign positive). Weakness in upper limbs was symmetrical and static. Patient also had difficulty in holding objects and falling of spoon was present while eating. There was no history of fever, vomiting, loose stool, headache and trauma.

At 2 year of age, child presented with failure to thrive. She was diagnosed with celiac disease based on raised IgA TTG levels (235 U/ml). The girl after starting gluten free diet started improving and showed significant weight and height gain. But after some years- parents stopped GFD and were giving normal home meals to child.

Anthropometry examination (IAP) showed weight/age <3rd centile, Height /age- <3rd centile and BMI-11.54 (<3rd centile). On physical examination, child was afebrile with vitals- Heart rate- 94 beats per minute, Respiratory rate- 18/min, SPO2- 97% on room air, Blood pressure- 105/70 mm Hg in left arm. Pallor present with no icterus/cyanosis/clubbing/edema/lymphadenopathy.

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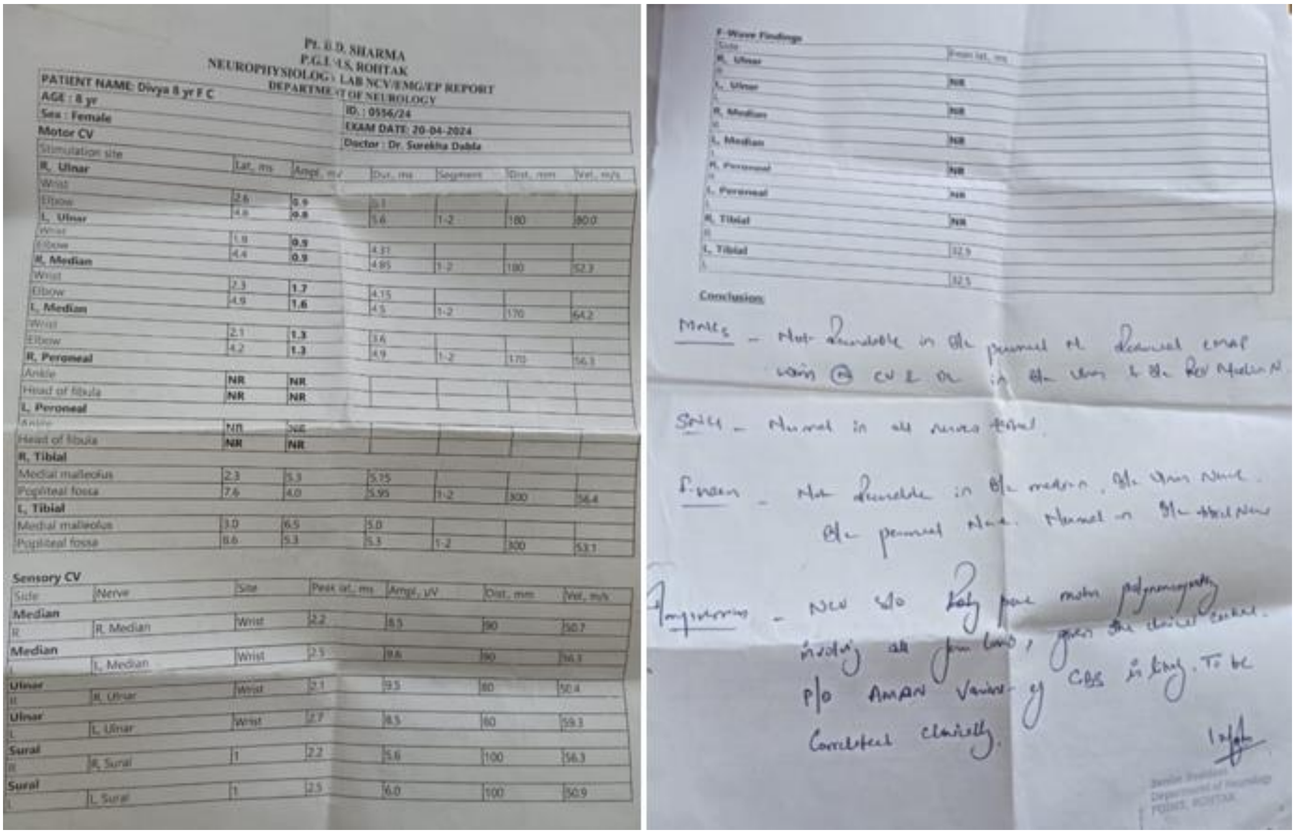

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NEUROPHYSIOLOGY LAB NCV/EMG/EP REPORT
DEPARTMENT OF NEUROLOGY

PATIENT NAME: Divya 8 yr F C
AGE : 8 yr
Sex : Female
ID. : 0556/24
EXAM DATE: 20-04-2024
Doctor : Dr. Surekha Dabla

Motor CV

Stimulation site	Lat, ms	Ampl, mV	Dur, ms	Segment	Dist, mm	Vel, m/s
R, Ulnar						
Wrist	2.6	0.9	5.1			
Elbow	4.8	0.8	5.6	1-2	180	80.0
L, Ulnar						
Wrist	1.8	0.9	4.31			
Elbow	4.4	0.9	4.85	1-2	180	52.3
R, Median						
Wrist	2.3	1.7	4.15			
Elbow	4.9	1.6	4.5	1-2	170	64.2
L, Median						
Wrist	2.1	1.3	3.6			
Elbow	4.2	1.3	4.9	1-2	170	56.3
R, Peroneal						
Ankle	NR	NR				
Head of fibula	NR	NR				
L, Peroneal						
Ankle	NR	NR				
Head of fibula	NR	NR				
R, Tibial						
Medial malleolus	2.3	5.3	5.75			
Popliteal fossa	7.6	4.0	5.95	1-2	300	56.4
L, Tibial						
Medial malleolus	3.0	6.5	5.0			
Popliteal fossa	8.6	5.3	5.3	1-2	300	53.1

Sensory CV

Side	Nerve	Site	Peak lat, ms	Ampl, µV	Dist, mm	Vel, m/s
Median						
R	R, Median	Wrist	2.2	8.5	90	50.7
Median						
L	L, Median	Wrist	2.5	9.6	90	56.1
Ulnar						
R	R, Ulnar	Wrist	2.1	9.5	80	50.4
Ulnar						
L	L, Ulnar	Wrist	2.7	8.5	80	59.3
Sural						
R	R, Sural	1	2.2	5.6	100	56.3
Sural						
L	L, Sural	1	2.5	6.0	100	50.9

F-Wave Findings

Side	Fmin lat, ms
R, Ulnar	
R	
L, Ulnar	NR
L	
R, Median	NR
R	
L, Median	NR
L	
R, Peroneal	NR
R	
L, Peroneal	NR
L	
R, Tibial	NR
R	
L, Tibial	32.9
L	32.5

Conclusion:

P/o AMAN Variety of GBS is likely. To be Correlated clinically.

Figure 1: Nerve conduction velocity suggestive of poly pure motor neuropathy (AMAN variety of GBS is likely).

Central nervous system examination showed, child was conscious and oriented to time, place and person. Tone normal in all limbs, power 4/5 in all limbs, DTR were absent. Gower sign was positive.

Investigation revealed Hb-10.8 gm/dl, wbc-11500/ mm^3 with neutrophils-51% & Lymphocyte- 42%, Platelet count- 2.7 lacs, CRP- negative, Peripheral smear-Normocytic Normochromic picture, blood urea- 24 mg/dl, serum Creatinine- 0.6mg/dl and Urine microscopy- normal. Repeat investigations showed Thyroid profile- normal, Serum IgA TTG- 280 U/ml, Vit B12 levels- 534 mg/dl. And CPK-MB=46 U/L (Range=0-24 U/L). Nerve conduction velocity showed pure motor polyneuropathy involving all 4 limbs. [**Figure 1**]

Differential diagnosis- Celiac disease associated neuropathy, Guillian Barre Syndrome, Nutritional deficiencies (Vit B12 deficiencies).

A probable diagnosis of celiac disease associated neuropathy was made. Point in favor of diagnosis- documentation with previously diagnosed celiac disease, elevated levels of serum IgA TTG, improvement in symptoms when started on gluten free diet, Nerve Conduction Velocity deciphering pure motor neuropathy. Girl was started on strict gluten free diet along with necessary supplements. She showed significant clinical improvement after 1 week and then was discharged with strict instructions for continuing gluten free diet along with calcium and multivitamin supplements. On her follow up visit at one month, the child had gained weight, was active, no Gower sign and neuropathy had improved. Parents consent was taken for publication purpose, as patient is minor.

3. Discussion

Peripheral neuropathy can be caused by a variety of systemic diseases, toxins, medications, infections and hereditary disorders.[4] Peripheral neuropathy associated with celiac disease is an atypical presentation. Gluten neuropathy is an autoimmune disorder in which gluten ingestion causes damage to nerves of the peripheral nervous system leading to disrupted communication from central nervous system to the rest of the body.[5]

The exact etiology of peripheral neuropathy associated with celiac disease is said to be idiopathic. Detailed review of literature points towards possibility of involvement of anti-ganglioside antibodies, toxic effects of Gliadin and associated multivitamin deficiencies in the pathogenesis of peripheral neuropathy associated with celiac disease.[6] Gluten neuropathy presents with variable severity. A retrospective study conducted on patients with celiac disease associated gluten neuropathy, disesase severity was categorized as mild in 27% (confined to legs), moderate in 40% (arms involved but radial nerve spared) and severe (33%, radial nerve involved).[7]

Index case presented with features of peripheral neuropathy and failure to thrive. Patient was diagnosed with celiac disease at 2 year of age, started on gluten free diet but stopped treatment after some time. Differential diagnosis included Guillian Barre syndrome, nutritional deficiencies and Peripheral neuropathy associated with celiac disease. Detailed investigative workup was conducted which excluded other probable causes like nutritional deficiencies, diabetes, hypothyroidism, vascular, infectious or toxic etiologies. Child was diagnosed with peripheral neuropathy associated with points in favour of diagnosis as following- previous documentation and history of celiac disease, elevated IgA TTG, improvement of symptoms on gluten free diet, NCV findings consistent with motor neuropathy. The child showed spontaneous recovery on strict gluten free diet. Follow up visits were done at intervals of 1 month and 3 month which showed significant improvement in growth and activity.

The interaction of gluten free diet on neurological disorder associated with celiac disease is unclear. Only very few literature mentioned complete neurological recovery after gluten free diet whereas other literature showed no recovery after gluten free diet.[8] But in our case, patient showed significant improvement after introduction of strict gluten free diet. There is a need of further studies to assess the effect of gluten free diet on these celiac associated disorders and to investigate the underlying mechanisms of nervous system involvement occurring due to gluten sensitivity.

4. Conclusion

Celiac disease associated peripheral neuropathy is an uncommon presentation and sometimes present as the only clinical feature without any GI manifestation. Celiac disease should be kept as probable diagnosis in all cases of peripheral neuropathy. Gluten free diet remains the cornerstone of treatment.

5. Source of Funding

None.

6. Conflict of Interest

None.

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