A CASE OF PROGRESSIVE **GAIT DISTURBANCE** AND **CHRONIC DELUSIONAL DISORDER** IN A CAUCASIAN 75-YEAR-OLD WOMAN WITH **OCCULT CELIAC DISEASE**

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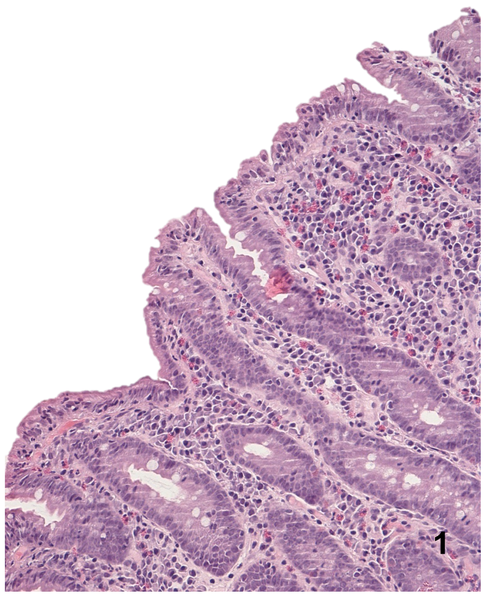


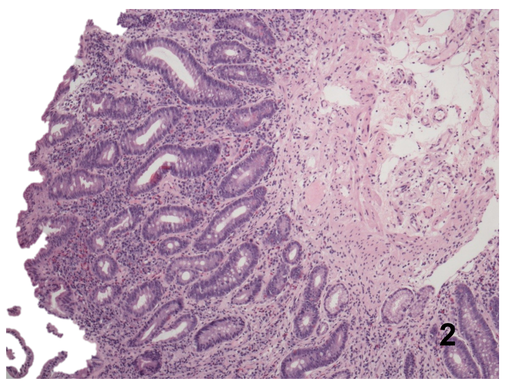


Fig. 1 e 2: Fragments of not oriented duodenal mucosa with total villous atrophy, glandular crypt hyperplasia (ratio villous/crypt altered), enterocytes of low height and low surface, brush-border irregularity, presence of cytoplasmic vacuoles.

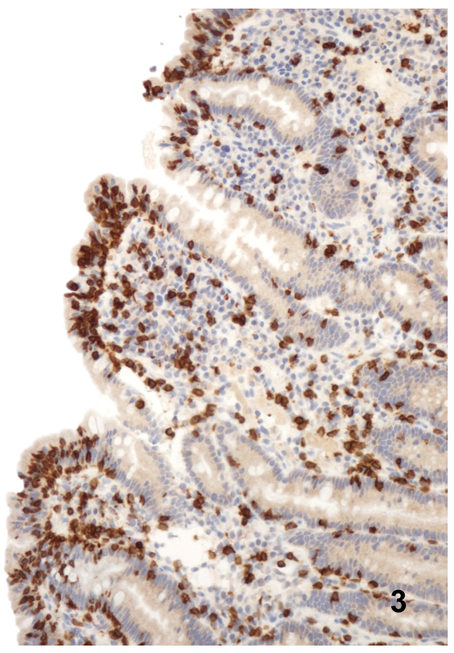


Fig. 3: Immunohistochemical staining for CD3 showed pathological increase in the percentage of intraepithelial lymphocytes (> 40/100 enterocytes)

The described findings are compatible with celiac disease with atrophic type lesions (lesion 3c according to Marsh mod Oberhuber or atrophic lesion GRADE B 2 according to Corazza / Villanacci)

Objective: to describe a challenging case of a complex neuro-psychiatric disorder, which after an extensive diagnostic workup, resulted to be caused by a silent celiac disease.

Background: CD is relatively frequent condition, and it could notoriously have an atypical clinical presentation with **extra-intestinal symptoms**. Neuropsychiatric disorders and CD are **not considered a simple random association**. However, the mechanisms involved in the pathophysiology of neurological and psychiatric disorders in CD are currently **unknown**

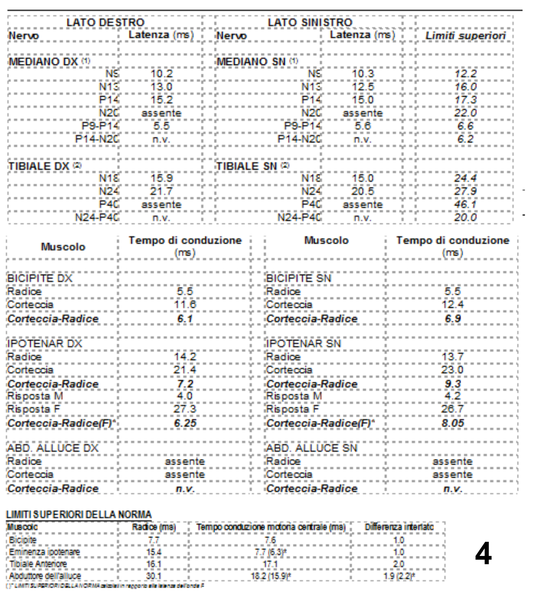
Case report: a **75-year old woman** presented with a **10-year** history of **progressive** gait disturbance characterized by a **deficit of strength** and **stiffness** sensation in both legs. Comorbidities included: a **chronic delusional disorder** (delusional jealousy) treated for about 7 years with aripiprazole 2.5mg/day; arterial hypertension; osteoporosis. Neurological evaluation revealed the simultaneous presence of **pyramidal signs** (severe spastic paraplegia with lower limbs hyper-reflexia and bilateral Babinski sign), **parkinsonism** (bradykinesia, rigidity, hypomimia, postural instability and resting tremor), a **cerebellar syndrome** (gait ataxia, bilateral distal kinetic tremor, cerebellar dysarthria) and signs of **polyneuropathy** (weakness and hypoesthesia in the distal region of the four limbs). Psychiatric evaluation confirmed the diagnosis of chronic delusional disorder.

We performed: -routine blood tests, normal except for a **severe folic acid deficiency** (0.70ng/mL n.v.>5.38); -brain MRI (bihemispheric white matter T2 hyperintensities and atrophy); -spine MRI (normal); -neurophysiologic assessment (EMG/NCS study, with findings suggestive of chronic axonal polyneuropathy with mild signs of acute denervation; evoked potentials, which confirmed a severe dysfunction of both central and peripheral motor and sensory pathways).

The finding of a severe folate deficiency on blood tests raised the suspicion of a possible malabsorption, therefore we tested: anti-transglutaminase and anti-endomysial antibodies (positive); homocysteine (normal); Vitamin B12 (normal but recently supplemented). A duodenal biopsy was performed, that led to the **histological confirmation of CD**. A **gluten-free diet** was started. At the 6-month and 15-month follow-up we observed a **subjective improvement** in the spasticity and fatigability of the lower limbs.

Discussion: Other possible differential diagnoses (Wilson diseases, MSA, SCA, HSP) were excluded. The delay in the diagnosis, mostly due to the absence of gastrointestinal manifestations, could have produced a **permanent neurological damage**.

Conclusions: CD has to be considered in the differential diagnosis of a broad spectrum of neurological and psychiatric diseases, even in the absence of gastrointestinal symptoms.



LATO DESTRO		LATO SINISTRO		
Nervo	Latenza (ms)	Nervo	Latenza (ms)	*Limiti superiori*
MEDIANO DX		MEDIANO SN		
N9	10.2	N9	10.3	12.2
N13	13.0	N13	12.5	16.0
P14	15.2	P14	15.0	17.3
N20	assente	N20	assente	22.0
P9-P14	5.5	P9-P14	5.6	6.6
P14-N20	n.v.	P14-N20	n.v.	6.2
TIBIALE DX		TIBIALE SN		
N18	15.9	N18	15.0	24.4
N24	21.7	N24	20.5	27.9
P40	assente	P40	assente	46.1
N24-P40	n.v.	N24-P40	n.v.	20.0

Muscolo	Tempo di conduzione (ms)	Muscolo	Tempo di conduzione (ms)
BICIPITE DX		BICIPITE SN	
Radice	5.5	Radice	5.5
Corteccia	11.6	Corteccia	12.4
Corteccia-Radice	***6.1***	***Corteccia-Radice***	***6.9***
IPOTENAR DX		IPOTENAR SN	
Radice	14.2	Radice	13.7
Corteccia	21.4	Corteccia	23.0
Corteccia-Radice	***7.2***	***Corteccia-Radice***	***9.3***
Risposta M	4.0	Risposta M	4.2
Risposta F	27.3	Risposta F	26.7
Corteccia-Radice(F)	***6.25***	***Corteccia-Radice(F)***	***8.05***
ABD. ALLUCE DX		ABD. ALLUCE SN	
Radice	assente	Radice	assente
Corteccia	assente	Corteccia	assente
Corteccia-Radice	***n.v.***	***Corteccia-Radice***	***n.v.***

LIMITI SUPERIORI DELLA NORMA

Muscolo	Radice (ms)	Tempo conduzione motoria centrale (ms)	Differenza interlato
Bicipite	7.7	7.6	1.0
Eminenza ipotenar	10.1	7.7 (6.9)*	1.0
Tibiale Anteriore	16.1	17.1	2.0
Adduttore dell'alluce	30.1	18.2 (15.9)*	1.9 (2.2)*

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Fig. 4: evoked potentials confirmed a severe dysfunction of both central and peripheral motor and sensory pathways (6months control)

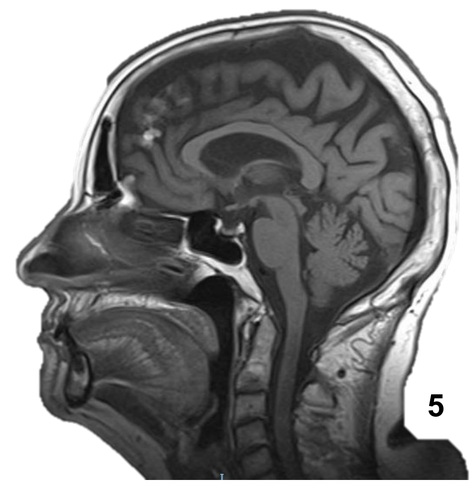

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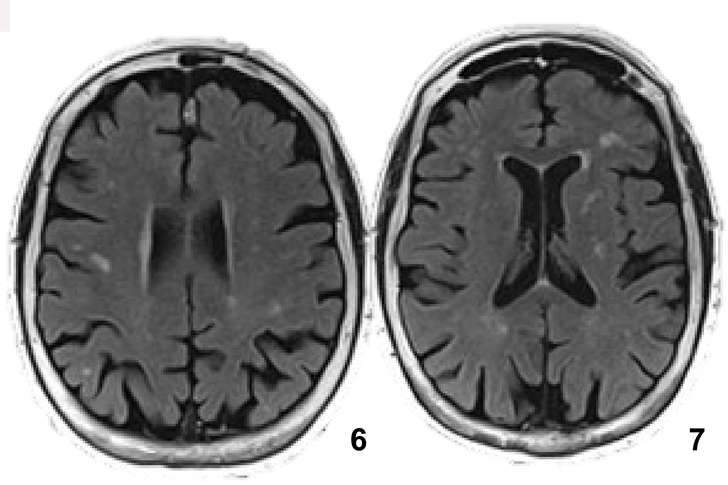
Fig. 5-11: brain MRI showed bi-hemispheric white matter T2 hyperintensities and atrophy; spine MRI was normal. *Note cerebellar atrophy*



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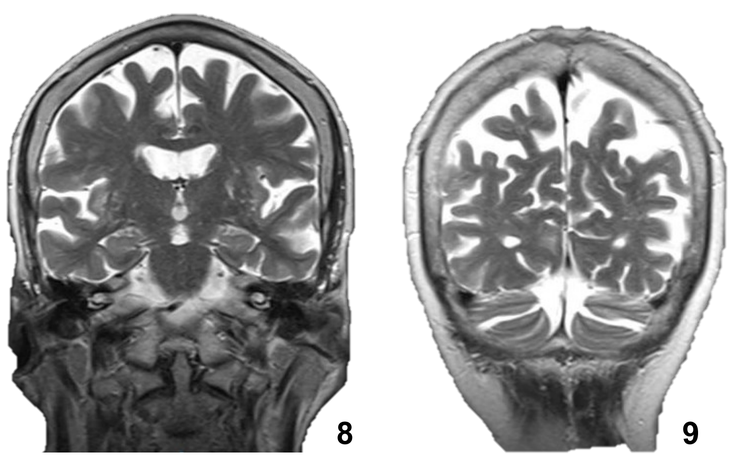

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