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A Case of Unicentric Castleman Disease Presenting with Unilateral Lumbosacral Radiculopathies

INTRODUCTION

Castleman disease, also known as angiofollicular lymph node hyperplasia, is a rare lymphoproliferative disorder characterized by benign proliferation of lymphoid tissue. Neurological manifestations in Castleman disease are rare, but can include neuropathies secondary to compression by enlarged lymph nodes or immune-mediated mechanisms. The case report addresses unicentric Castleman disease presenting with unilateral lower extremity weakness as an initial symptom.

CASE PRESENTATION

Present illness & Physical Examination

- A 55-year-old male patient
- Sensory change, progressive weakness, and subjective muscle atrophy in his left lower extremity over the past three years
- hypoesthesia on the dorsum and sole of the left foot, along with mild hypoesthesia in the lateral aspect of the left leg and buttock,
- muscle strength of the left lower extremity
hip extension and abduction, 3; knee flexion and ankle plantarflexion 4; ankle dorsiflexion and great toe extension, 0 ~ 1
- muscle atrophy in the thigh and calf, along with a hypoactive ankle reflex on the left side

ELECTROPHYSIOLOGICAL, RADIOLOGICAL, & HISTOLOGICAL FINDINGS

NERVE CONDUCTION STUDIES

Side	Motor Nerve	Stimulation	Recording	Latency (msec)		Amplitude (mV)	NCV (m/s)	F wave (msec)
Lt.	Tibial	ankle	AH	5.3	6.6*	36*		62.1*
		popliteal fossa		14.9	4.3			
	Peroneal	ankle	EDB		0			NR*
	Peroneal	ankle	TA		0			
	Medial plantar	ankle	FHB	5.9	5.3			
	Inferior calcaneal	ankle	ADM	5.9	3.0*			
	Lateral plantar	ankle	FDMB	6.2	2.5*			
Rt.	Lateral plantar	MM	FDI	7.0	2.8*			
		MM-5		5.8	2.9			
	Tibial	ankle	AH	4.2	13.5			
	Peroneal	ankle	TA	2.6	7.9	50		
		fibular head		3.6	7.6			
	Medial plantar	ankle	FHB	5.2	7.5			
	Inferior calcaneal	ankle	ADM	5.2	9.0			
	Lateral plantar	ankle	FDMB	5.7	5.1			
	Lateral plantar	MM	FDI	6.1	8.2	63		
		MM-5		5.3	8.5			
Side	Sensory Nerve	Stimulation	Recording	Latency (msec)		Amplitude (uV)	Distance (cm)	
				Onset	Peak			
Lt.	Sural	calf	ankle			NR*	14	
	Superficial peroneal	calf	ankle			NR*	14	
	Medial plantar	sole	ankle			NR*	14	
	Lateral plantar	ankle	sole			NR*	14	
Rt.	Sural	calf	ankle	2.3	3.1	11	14	
	Superficial peroneal	calf	ankle	3.4	4.1	5	14	
	Medial plantar	sole	ankle	2.6	3.2	3	14	
	Lateral plantar	ankle	sole	2.7	3.2	3	14	
Soleus H-reflex		Rt.	Lt.					
Latency (msec)		33.6*	NR*					

NEEDLE ELECTROMYOGRAPHIC EXAMINATION

Muscle	Insert. activity	Fib/PSW	Motor Unit Action Potentials				Recruitment
			Normal	Configuration	Amplitude	Duration	
Lt.	Iliopsoas	N	-	N			F
	Adductor longus	N	-	N			F
	Vastus lateralis	N	-	N			F
	Tibialis anterior		F&P (++)				No M.U.A.P
	Peroneus longus		F&P (++)				No M.U.A.P
	Gastrocnemius med.		F&P (++)	Polys		Long	R
	Biceps femoris, short head		F&P (+)	Complex polys			D
	Medial hamstrings		tiny F&P(+)	Polys			max R
	Flexor digitorum longus		F&P (+)	Polys		Long	R
	Flexor hallucis brevis		F&P (+)	Polys	Large(10mV)	Long	R
	Gluteus maximus (upper portion)		tiny F&P (++++)				No M.U.A.P
	Gluteus maximus (lower portion)		tiny F&P (++++)				S
	Tensor fascia latae	N	-	N			F
	Gluteus medius	N	-	N			F
	External anal sphincter	N	-	N			F
	L4/5 PVM	N	-				
	L5/S1 PVM	N	-				
	S1/S2 PVM		F&P (+)				
	S2/S3 PVM		F&P (+)				
	S3/S4 PVM		F&P (+)				

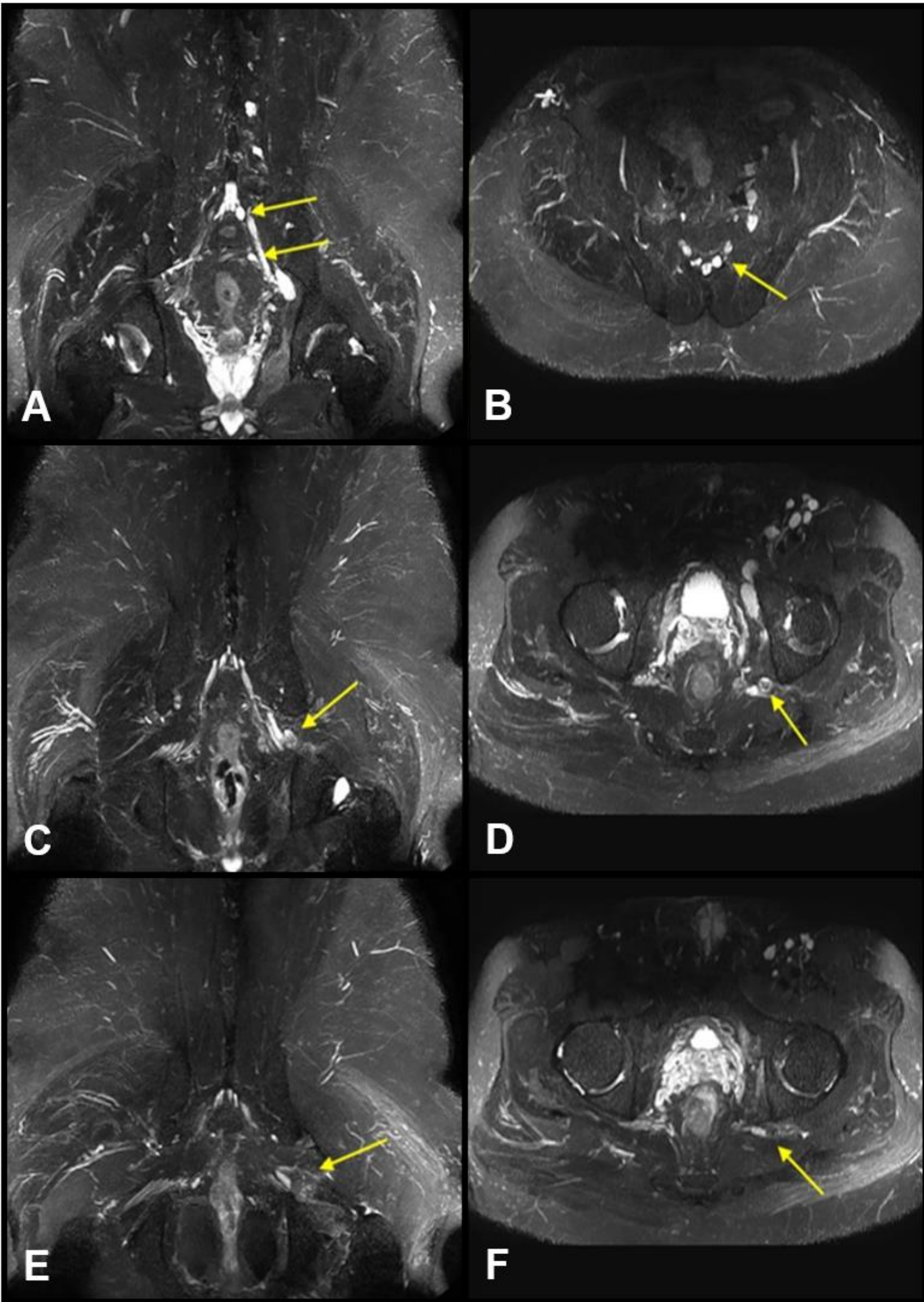


Fig. 1. MRI demonstrates coronal and axial images of lumbar and sacral plexus. T2 hyperintensity with marked swelling of the left S1-2 nerve roots (A,B) and left proximal sciatic nerve are noted (C,D), with enhancing perineural infiltration of the sciatic nerve posterior to the posterior acetabulum (E,F), as indicated by the yellow arrows.

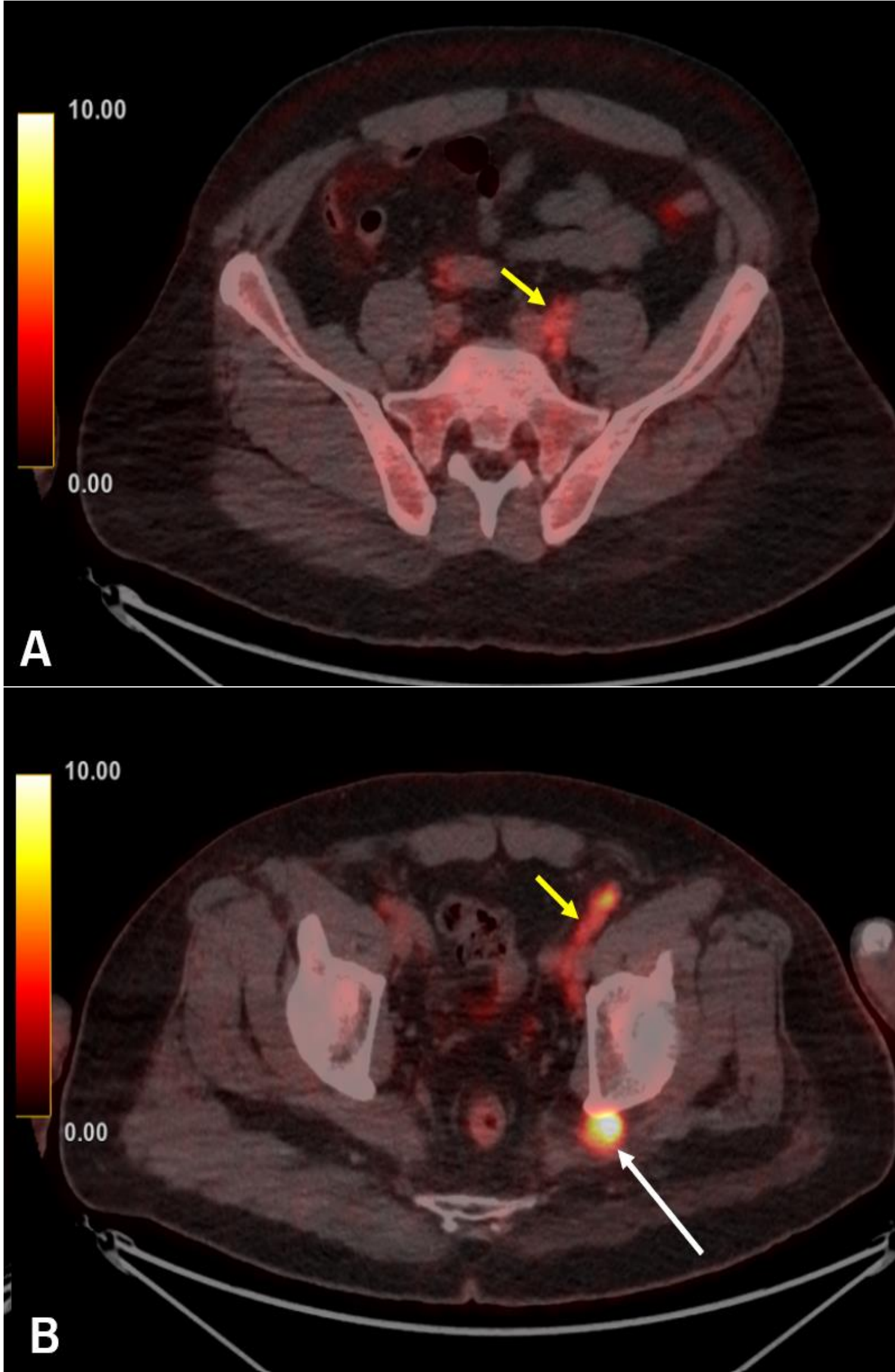


Fig. 2. Torso PET-CT (FDG) demonstrates multiple hypermetabolic lymph nodes along the left ilioinguinal chain (A, B), as indicated by the yellow arrows, and a hypermetabolic lesion posterior to the left posterior acetabulum (B), indicated by the white arrow, suggesting sciatic nerve involvement.

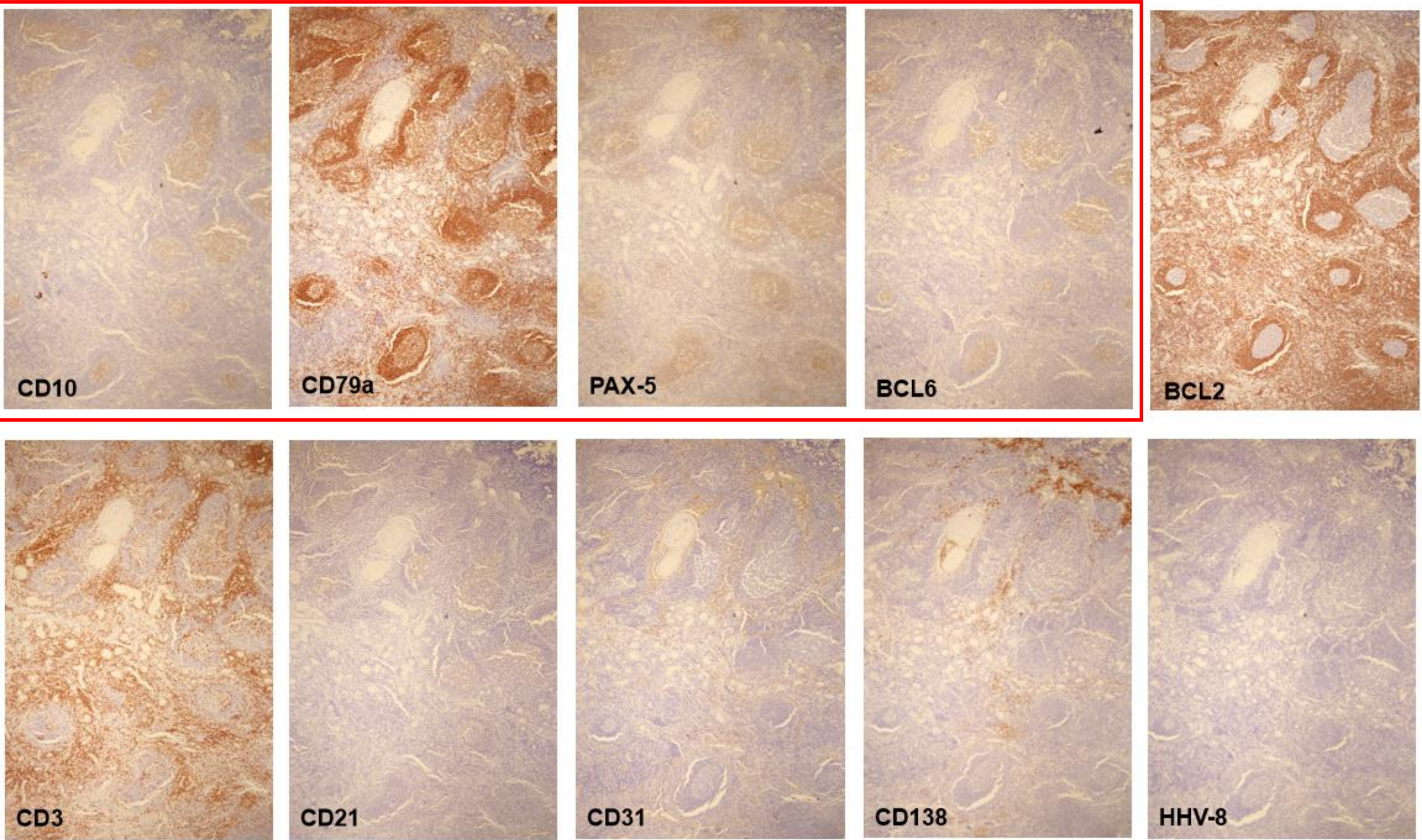


Fig. 3. excisional biopsy of a left inguinal lymph node and subsequent immunohistochemical staining. POSITIVE: CD10, CD20, CD79a, FAX-5,BCL-6 NEGATIVE: CD3, CD5, CD21, CD31, CD34, CD138, BCL-2, HHV-8, MUM-1, Cyclin D1,

CONCLUSION

This is the first case report of unicentric Castleman disease presenting with unilateral lumbosacral radiculoplexopathy. In Castleman disease presenting with atypical symptoms and signs, diagnostic imaging modalities, such as MRI and PET-CT, play a crucial role in diagnosis. To confirm Castleman disease, a lymph node biopsy has to be performed. Integrating these imaging findings with clinical and histopathological data is crucial for an accurate diagnosis and tailored management of this complex disease entity.