

Case Report : Intrathecal baclofen use in stiff person syndrome

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Background:

- **Rare Autoimmune Disorder:** Progressive muscle stiffness, painful spasms, and walking difficulties; often linked to anti-GAD65 antibodies.
- **Diagnostic Challenge:** Easily misdiagnosed due to overlap with other neuromuscular conditions.
- **Limitations of Oral Therapy:** Baclofen, diazepam, and tizanidine may give partial relief and cause side effects.
- **Intrathecal Baclofen (ITB):** Effective in refractory cases, showing significant symptom improvement.

Case:

Patient Background

- 57-year-old male with right auricular melanoma
- Previous treatments: partial auriculectomy, sentinel node biopsy, radiotherapy, pembrolizumab
- 4 years later: fatigue, dizziness, hand tremors

Disease Progression and Imaging

- Lung nodule → metastatic melanoma confirmed
- PET scan: hypermetabolic activity (lung, subcarinal & cervical nodes, adrenal gland)
- Labs: eosinophilia
- EMG: lower extremity motor neuron/axon injury

Symptom Management (Pre-Diagnosis)

- Symptoms: generalized pain, cramps, dysphagia, fatigue, disrupted sleep
- Medications tried: baclofen, tizanidine, hydrocodone, diazepam, gabapentin, tramadol, prednisone, dantrolene
- Partial relief: diazepam (cramps), gabapentin (neuropathic pain)
- Physical therapy: aquatic therapy unsuccessful
- Oswestry Disability Index (from 0-100): 28 (back; Moderate Disability), 26 (neck; Moderate Disability)

Diagnosis and Targeted Therapy

- Referred to pain management → **diagnosed with Stiff Person Syndrome**
- Intrathecal baclofen (ITB) pump implanted:
 - Start: 75 mcg/day → Titrated to 196 mcg/day
- Outcomes:
 - Complete e resolution of lower extremity stiffness
 - Reduced pain & opioid requirement

Current Status and Ongoing Issues

- Persistent stiffness & pain: upper extremities, neck, jaw
- Supportive care: stool softeners, ongoing monitoring
- Opioid Risk Tool & CAGE-AID: negative

Discussion:

- **Intrathecal baclofen (ITB)** offers targeted GABAergic modulation, improving stiffness and spasms in refractory SPS while minimizing systemic side effects (Dalakas, 2009; McKeon et al., 2012; Dalakas, 2020).
- **Multidisciplinary management**—involving neurology, pain medicine, and physical therapy—is crucial for optimizing outcomes and maintaining functional improvement (Murinson & Guarnaccia, 2008; Rakocevic & Floeter, 2016)

Conclusion:

- **Intrathecal baclofen pump therapy** is a safe and effective option for patients with refractory stiff person syndrome, providing marked symptom relief and improved quality of life.
- **Early diagnosis and multidisciplinary management** are essential for optimizing outcomes in this rare and disabling neurological disorder.

References:

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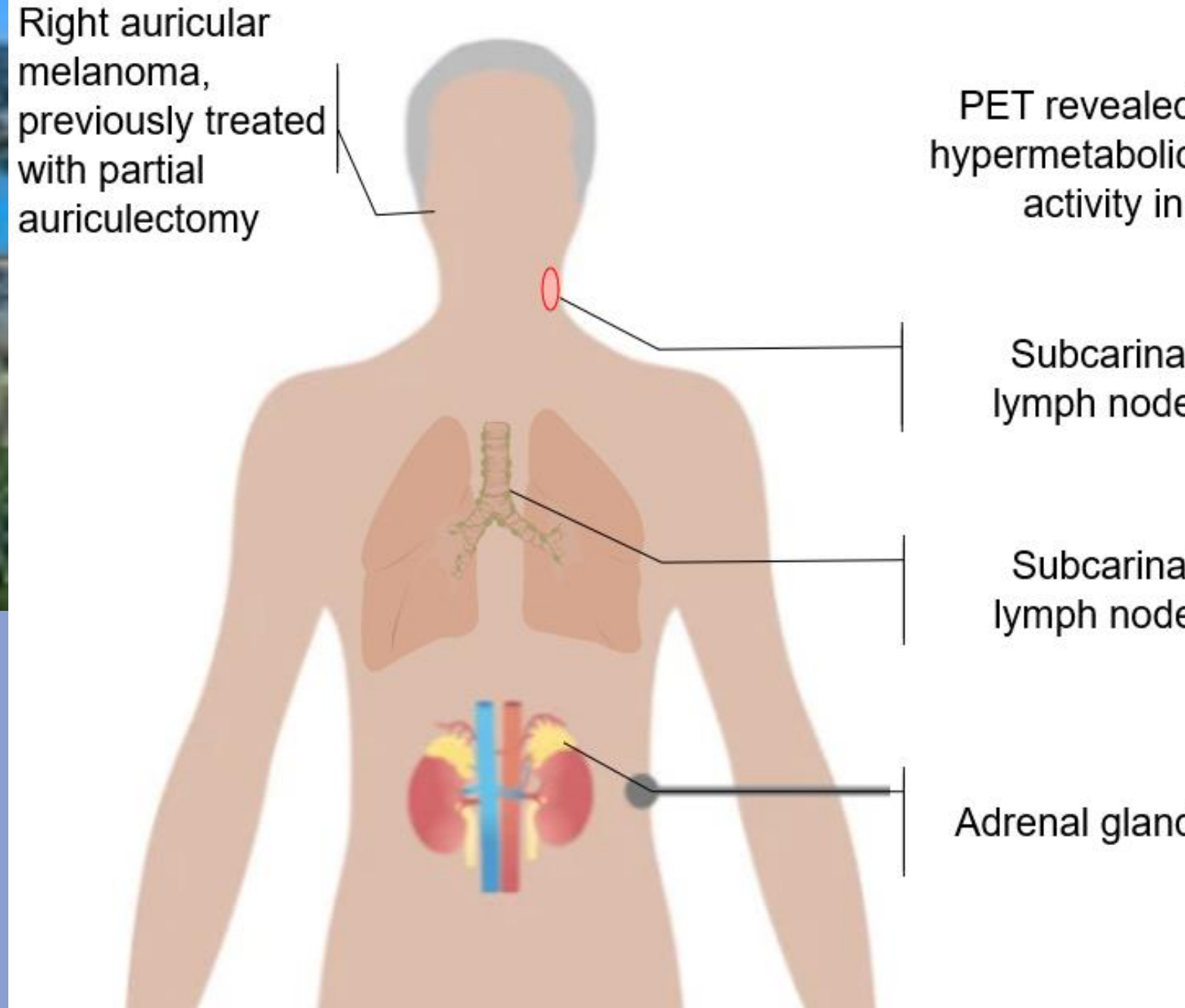


Figure 1: Clinical presentation of the case at first clinic visit.

Table 1 : Medications Tried and Clinical Response

Medication / Therapy	Response	Notes / Reason for Discontinuation
Baclofen (oral)	⚠ Limited relief	Incomplete symptom control
Diazepam	✅ Effective	Improved muscle cramps, reduced spasms
Tizanidine	⚠ Partial relief	Some benefit after reintroduction
Gabapentin	✅ Effective	Reduced neuropathic pain
Prednisone (5 mg daily)	⚠ Partial relief	Helped with inflammation and pain
Hydrocodone/Acetaminophen	⚠ Partial relief	Pain reduced after ITB pump
Tramadol	❌ Ineffective	Minimal pain control
Dantrolene (Dantrium)	❌ Ineffective / Stopped	Side effects: insomnia, dysgeusia, polyuria
Aquatic Physical Therapy	❌ Ineffective	Discontinued after two sessions
Intrathecal Baclofen (ITB)	✅ Highly effective	Marked improvement in stiffness & pain

Figure 3: Clinical timeline of a 57-year-old male with metastatic melanoma and refractory Stiff Person Syndrome, showing limited response to oral agents. Intrathecal baclofen pump initiated at 75 mcg/day and titrated to 196 mcg/day, resulting in marked reduction in stiffness and pain with improved lower extremity mobility.

