Background
• Intrathecal baclofen (ITB) drug-related complications from overdose or withdrawal are potentially life-threatening requiring prompt and accurate diagnosis and treatment.
• Symptoms of ITB withdrawal include:
  • Rebound spasticity
  • Pruritus
  • Hyperthermia
  • Diaphoresis
  • Seizures
  • Priapism
• ITB has been observed to have inhibitory effect on erectile function
• Urologists have successfully treated priapism with oral and intrathecal baclofen.
• No cases of priapism as a presenting symptom of ITB withdrawal have been described.

Study Design & Participants
• Retrospective case series
• Four males with cerebral palsy (CP) who presented to our institution with signs of acute ITB withdrawal, including increased tone and priapism

Case Descriptions

Case 1: 8 y/o with quadriplegic CP underwent revision of fractured catheter. Pre-operative concerns of increased tone for 3 months despite 183mcg/day ITB and phenol injections. Post-operative ITB dose started at 96mcg/day. Post-operative course complicated by priapism felt to be due to ITB withdrawal due to lower dose. Dose rapidly increased to baseline with resolution of priapism.

Case 2: 17 y/o with quadriplegic CP presented to clinic with an alarming ITB pump and priapism. Pump interrogation revealed pump alarming due to low reservoir. ITB pump was refilled confirming residual volume of 0mL. 25mcg bolus administered and pump started at previous dose. Priapism resolved with these interventions.

Case 3: 18 y/o with diplegic CP presented to clinic with complaints of ITB pump alarming every 10 minutes since one day prior, painful priapism, increased tone, and pruritus. Pump interrogation revealed motor stall had occurred. The pump was reprogrammed but motor stall remained. Patient was started on oral baclofen 20mg q 6 hours. Priapism resolved with oral baclofen. The patient underwent ITB pump replacement.

Case 4: 19 y/o with quadriplegic CP presented to clinic with one day history of increased tone, difficulty sleeping, and priapism. Pump series x-rays revealed two areas of possible catheter kinking. Pump catheter access attempted but no fluid withdrawn. The patient had a history of his pump flipping in the pocket. His pump and catheter were repositioned and priapism resolved with re-initiation of therapy.

Results
• In all four cases priapism resolved with re-initiation of baclofen delivery

Significance
• Although priapism has been mentioned as a clinical feature of ITB withdrawal there are no cases reported in the literature.
• Including priapism as one of the symptoms of ITB withdrawal may assist with the early recognition of this potentially life-threatening condition.

Table 1. Baclofen Use in the Treatment of Priapism

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Presentation</th>
<th>Treatment</th>
<th>Outcomes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bourke. 2002 J of Urology</td>
<td>2</td>
<td>Recurrent idiopathic nocturnal priapism</td>
<td>PO Baclofen 40mg q hs</td>
<td>No priapism at 5-12 mo</td>
</tr>
<tr>
<td>Vaidyanathan 2004 Spinal Cord</td>
<td>1</td>
<td>Priapism elicited by movement in C4 SCI</td>
<td>PO Baclofen 10mg TID</td>
<td>Erections not triggered by movement</td>
</tr>
<tr>
<td>Moreira 2006 J Pediatr Urol</td>
<td>1</td>
<td>Idiopathic recurrent priapism</td>
<td>PO Baclofen 30mg q hs</td>
<td>No recurrence at 6 mo</td>
</tr>
<tr>
<td>Van Driel 2008 J Sex Med</td>
<td>1</td>
<td>Sleep-related painful erection</td>
<td>PO Baclofen 30mg q hs</td>
<td>Pt satisfied at 4 mo</td>
</tr>
<tr>
<td>D’Aleo 2009 Spine</td>
<td>1</td>
<td>Priapism in C5 complete SCI</td>
<td>PO Baclofen 75mg q day ITB 180mcg/day</td>
<td>Absence of priapism with ITB</td>
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