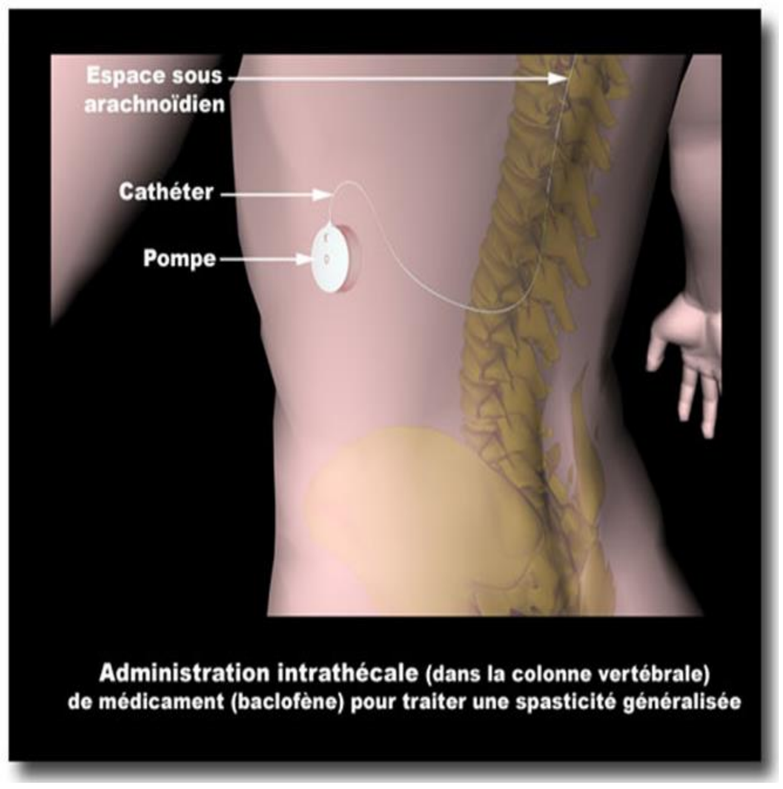
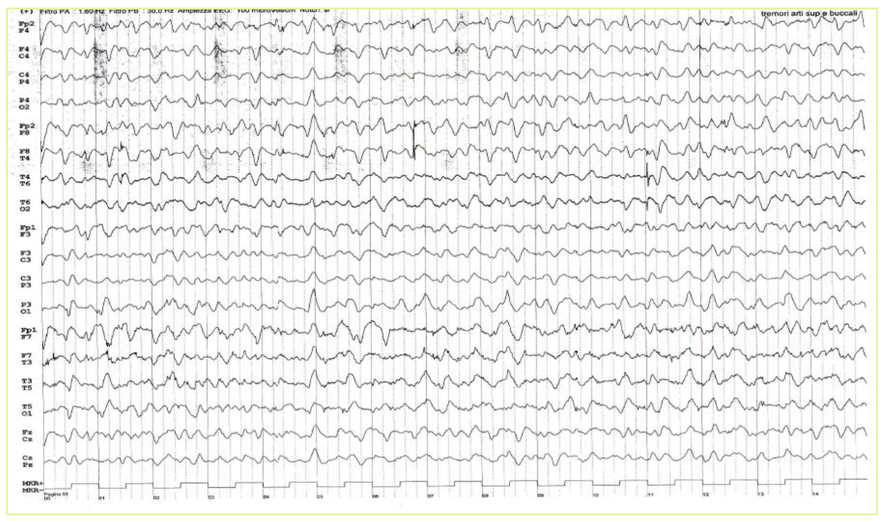
Intrathecal Baclofen Withdrawal: unusual symptomatic presentation and successful treatment

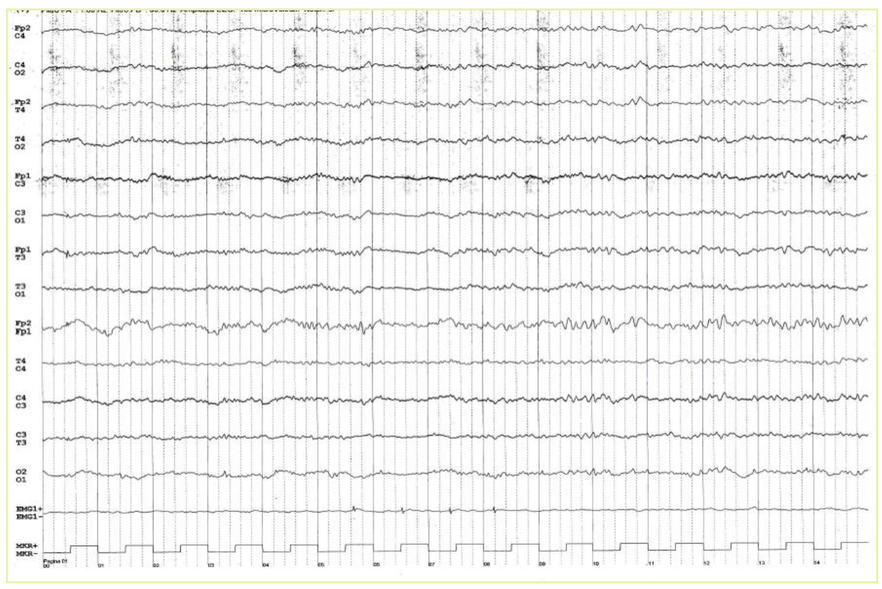
M. Di Stefano, P. Melzi, L. Airoldi, G. Costantino, E. Tagliabue, A. Tetto, M. Vaccaro, A. Salmaggi
Department of Neurology, San Leopoldo Mandic Hospital, Merate (Lecco)

Intrathecal baclofen therapy is widely used to alleviate intractable spasticity through its gamma-Aminobutyric acid-B (GABA B) agonist properties.
The pump is implanted in the lower abdomen and dispenses medication from its reservoir through a silicon catheter into the thoraco-lumbar region intrathecally.
Complications like overdose or withdrawal can occur and could be the result of pump malfunction or refilling and programming mistakes.
Baclofen intoxication is due to pump refilling and programming mistakes and generally occurs in the first two days. It can lead to muscular hypotonia, generalized tonicoclonic seizures, respiratory depression, altered mental status and coma. Baclofen withdrawal can lead to perspiration, autonomic dysreflexia, rigidity, seizures, hyperthermia, stupor and coma.
Withdrawal mimicking sepsis and hypotension has been reported in several publications. Severe cases may present with rhabdomyolysis, multiorgan dysfunction and death.



A 41 year old patient with hereditary spastic paraparesis and intrathecal baclofen pump device presented sopor, autonomic dysreflexia, rigidity, hypothermia, arterial hypotension and respiratory insufficiency. Electroencephalography documented diffuse intermittent rhythmic delta activity (IRDA). A cerebral CT scan excluded acute intracranial lesion. Cerebrospinal fluid analysis was normal for cells, protein and glucose.
Our patient was towards the end of charge pump, and pump refilling was scheduled one week after onset of symptoms after the usual 6-month period. The patient was intubated and treated with midazolam (5 mg/h) and dopamine (5 mcg/Kg/min) infusion in intensive care unit for vital signs close monitoring. Intrathecal baclofen therapy was stopped. The patient was placed on enteral baclofen (12.5 mg, every 8 h) via nasogastric tube. Midazolam infusion was stopped approximately 58 h after. The patient was successfully extubated approximately 96 h after.
The electroencephalogram showed normal awake architecture and regular background activity. He was discharged home 10 days after admission with complete resolution of the autonomic changes and altered mental status.





Baclofen withdrawal is a life threatening condition that may be associated with reflex spasticity, dysautonomia, hyperthermia, depressed sensorium, multiorgan dysfunction, disseminated intravascular coagulopathy and death.
Benzodiazepines and enteral baclofen are the mainstay of treatment for acute intrathecal baclofen withdrawal.
We report a case of safe and efficacious use of midazolam infusion and enteral baclofen in a patient with hereditary spastic paraparesis and intrathecal baclofen pump device towards the end of charge pump.
The therapeutic approach was based on clinical criteria that considered especially the timing of onset of symptoms since the last pump refilling.
Hyperthermia was an unusual and abashing symptom of baclofen withdrawal. Symptomatic presentation is similar to alcohol withdrawal, which is mediated by modification of GABA A expression.
Benzodiazepines and baclofen administration induced progressive haemodynamic stability and complete resolution of the autonomic dysreflexia and altered mental status.

References
Albright AL, et al. Long-term intrathecal baclofen therapy for severe spasticità of cerebral origin. J.Neurosurg. 2003, 98, 291-5
Kao LW, et al. Intrathecal baclofen withdrawal mimicking sepsis. J.Emerg.Med. 2003, 24, 423-7
Coffey RJ, et al. Abrupt withdrawal from intrathecal baclofen: recognition and management of a potentially life-threatening syndrome. Arch.Phys.Med.Rehabil. 2002, 83, 735-41
Douglas AF, et al. Prolonged intrathecal baclofen withdrawal syndrome. Case report and discussion of current therapeutic management. J Neurosurg 2005; 102: 1133-6

Sin
XLVI CONGRESSO NAZIONALE
10-13 OTTOBRE 2015 – GENOVA
WebPoster