

# Pre- and postnatal botulinum toxin abdominal wall muscle relaxation in hepato-omphalocele

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## ABSTRACT

**Introduction.** Giant omphalocele poses a conflict between eviscerated content and abdominal capacity, with associated risks such as compartment syndrome or cardiovascular compromise.

**Clinical case.** We present the case of a prenatally diagnosed hepato-omphalocele, without associated abnormalities. At week 37, botulinum toxin was injected in the right hemiabdomen under fetal and maternal sedation. Following Cesarean section, scheduled at week 39, injection was completed in the left hemiabdomen, and full hepatic content reduction and total amnion inversion were achieved. Definitive repair with component separation was carried out on day 8 postnatally, and the patient was discharged on day 14.

**Discussion.** Botulinum toxin induces muscle relaxation, which can help reintroduce the eviscerated content within the abdominal cavity. The maximum effect is achieved following two weeks, which means prenatal injection may favor early amnion inversion, thus reducing hospital stay.

**KEY WORDS:** Omphalocele; Botulinum toxin; Fetal therapy.

## MIORRELAJACIÓN PRE Y POSTNATAL DE PARED ABDOMINAL CON TOXINA BOTULÍNICA EN HEPATOONFALOCELE

### RESUMEN

**Introducción.** Los onfalocelos gigantes plantean un conflicto entre el contenido eviscerado y la capacidad abdominal, con riesgos asociados como son el síndrome compartimental o el compromiso cardiovascular.

**Caso clínico.** Presentamos el caso de un hepatoonfalocelo con diagnóstico prenatal, sin anomalías asociadas. En semana 37 bajo sedación materna y fetal se realizó inyección de toxina botulínica en el hemiabdomen derecho. Tras la cesárea programada en semana 39 se completó inyección en el hemiabdomen izquierdo y se logró reducción completa del contenido hepático e inversión total del amnios. Se realizó reparación definitiva con separación de componentes el octavo día de vida y alta el decimocuarto día de vida.

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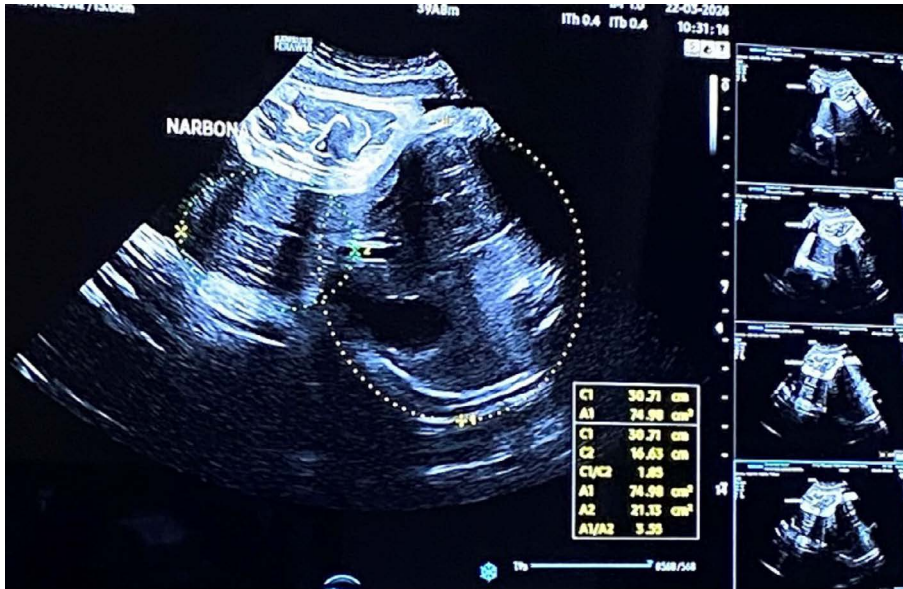
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**Comentarios.** La toxina botulínica provoca una miorelajación que puede favorecer la reintroducción del contenido eviscerado en cavidad abdominal. Su efecto máximo se alcanza a las dos semanas, por lo que su inyección en el período prenatal puede favorecer una inversión del amnios precoz, disminuyendo así la estancia hospitalaria.

**PALABRAS CLAVE:** Onfalocelo; Toxina botulínica; Terapia fetal.

## INTRODUCTION

Anterior wall defects represent a large pathological spectrum in pediatric surgery. Gastroschisis and omphalocele (exomphalos) are typically defined as such, with the latter being regarded as small in the presence of < 5 cm defects, and as giant in the case of > 5 cm defects and/or > 50% liver involvement<sup>(1)</sup>. This entity is associated with higher morbidity and mortality rates, not only as a result of the malformations associated (pulmonary hypertension, cardiac abnormalities, genetic disorders such as Beckwith- Wiedemann syndrome, pentalogy of Cantrell, or trisomies 13 and 18) in 30-80% of the cases<sup>(2)</sup>, but also because content reintroduction within the abdomen may entail increased intra-abdominal pressure, cardiopulmonary compromise, and hepatic vessel angulation. To repair such defects, a step-by-step closure is conducted by creating a surgical or non-surgical silo (silo of Abello)<sup>(3)</sup>, or by resorting to vacuum therapy, tissue expanders, or amnion epithelization and delayed closure. Botulinum toxin abdominal muscle relaxation has been described in adult and pediatric patients<sup>(4)</sup> in an attempt to increase abdominal capacity and reduce organ-abdominal disproportion in large ventral hernia and giant omphalocele. Abdominal capacity has been reported to grow by 25-30%, with infiltration in the transversus abdominis plane (TAP)<sup>(5)</sup> at the subcostal (9<sup>th</sup> rib), lateral (10<sup>th</sup>-12<sup>th</sup> rib), and anterosuperior iliac spine level. Even though the maximum effect of botulinum toxin is achieved following approximately two weeks, postnatal infiltration may play a part in the management of these



**Figure 1.** Prenatal ultrasonography: giant hepato-omphalocele.

patients. However, we suggest infiltrating botulinum toxin prenatally<sup>(6)</sup> so that the maximum effect is achieved at birth, thus optimizing abdominal wall relaxation.

## CLINICAL CASE

We present the case of a prenatally (week 14) diagnosed giant omphalocele. Following baseline diagnosis, chromosomal abnormalities were ruled out. The Pediatric Surgery Department was contacted by the Gynecology and Obstetrics Department, where prenatal diagnosis had been achieved, for prenatal assessment purposes. Prenatal botulinum toxin infiltration was considered.

Botulinum toxin infiltration in the fetal transversus abdominis plane (TAP) was scheduled at week 37. The procedure was conducted by a cross-disciplinary team of gynecologists, pediatric surgeons, neonatologists, and anesthesiologists, under maternal sedation. Fetal sedation was carried out by means of an intramuscular injection of atropine, fentanyl, and rocuronium. Given the intra-uterine position of the fetus, only the right transversus abdominis plane could be infiltrated. With a single placenta infiltration, botulinum toxin (12 IU/kg dose) was injected at the subcostal, 10<sup>th</sup> rib, and right iliac fossa level. No complications were recorded during the process or in the following days.

Cesarean section was scheduled at week 39. At birth, a plastic bag silo was placed on the defect. Once the patient had been relaxed and intubated, botulinum toxin was infiltrated on the left side, and a relaxation test was conducted, with full reduction of the eviscerated content and amnion inversion on the first day of life. Intra-abdominal pressure (IAP) was constantly monitored through bladder cath-



**Figure 2.** Botulinum toxin infiltration sites.

terization and INVOS renal flow control. A hydrocolloid dressing (Varihesive Gel Control™) was placed on the defect and changed every 48 hours. Trophic nutrition was initiated on the third day of life, and the patient was uneventfully extubated on the fourth, which allowed breastfeeding to be started.

Definitive wall closure was scheduled on the eighth day of life. The Ramírez component separation technique<sup>(7)</sup>, which involves performing an incision on the semilunar line to detach the rectus muscle from the external oblique muscle, was used. At the end of the procedure, intra-abdominal pressure was 10, and renal flow was adequate.

The patient remained intubated in the first 24 hours postoperatively. Tolerance was initiated on the first postoperative day, with progression being favorable, which led to discharge on day 14 postnatally.



**Figure 3.** Amnion inversion on the first day of life. Defect covered with a hydrocolloid dressing.

## DISCUSSION

The management of giant omphalocele represents a challenge for pediatric surgeons, and is currently not standardized. Botulinum toxin acts locally by blocking acetylcholine release<sup>(8)</sup>, which induces temporary muscle paralysis. When diagnosis has been achieved prenatally, we suggest infiltration is conducted at around week 37 –in the absence of contraindications. On the one hand, this favors abdominal muscle relaxation and therefore leads to less intrauterine abdominal content herniation, and on the other, it facilitates content reduction at birth, thus making mechanical ventilation shorter, which proves beneficial for the patient. The non-surgical (Abello) silo allows for a gradual reduction of the intestinal content until amnion inversion is reached. At this point, tolerance can be initiated so that intestinal peristalsis also favors abdominal compliance. This also allows breastfeeding to be reintroduced earlier, with the resulting benefit for the patient. Definitive repair –either by direct closure (if possible), by mobilizing the anterior sheath of the rectus muscles (San Martín technique)<sup>(9)</sup>, by placing surgical meshes, or using the Ramírez component separation technique– can therefore be scheduled.



**Figure 4.** First postoperative day following component separation.

Botulinum toxin therapy should be considered in the management of multiple pediatric pathologies. In the case of giant omphalocele, it allows for abdominal muscle relaxation, which helps reintroduce the abdominal content. Given that the maximum effect is achieved two weeks following infiltration, we suggest the latter is conducted prenatally in the presence of prenatal diagnosis and in the absence of contraindications.

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