Case report: incomplete Cauda Equina Syndrome following a caesarean section with spinal anaesthesia

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ABSTRACT

Background. The Cauda Equina Syndrome is a condition of severe compression or inflammation of the nerves that make up the cauda, that is the anatomical structure consisting of the last spinal roots [1].

Case presentation. On the second day after a caesarean section, a 34-year-old woman at her third pregnancy referred urinary retention, dysesthesia in the buttock and posterior region of the left thigh; constipation on the fifth day. After several clinical examinations, and having excluded the more statistically likely aetiologies, an incomplete Cauda Equina Syndrome was diagnosed, caused by chemical arachnoiditis from a spinal anaesthesia based on hyperbaric bupivacaine. The pathology resolved spontaneously and was not necessary to adopt any therapy other than the minimally invasive one. The urological symptoms were the first to completely disappear, while constipation resolved a few weeks later. Today, only a slight dysesthesia remains in the aforementioned areas.

Conclusions. Spinal anaesthesia still proves to be a safe and absolutely practicable method, even if not totally free of serious risks.

Key words
Caesarean-section; bupivacaine; spinal-anesthesia; Cauda-Equina syndrome; neurological-disease; urinary-retention.

INTRODUCTION

The Cauda Equina Syndrome is a condition of severe compression or inflammation of the nerves that make up the cauda, that is the anatomical structure consisting of the last spinal roots (L2-S5 and coccygeal level) [1].

It is characterised by five signs and symptoms, not always all present, namely: bilateral neurogenic sciatica, a reduction of perineal sensitivity, alteration of the bladder function, which can reach urinary retention without perception of urination stimulus, changes in anal tonality and sexual dysfunction [2].

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The causes, in a healthy patient who undergoes a caesarean section, can be varied and all related to neuraxial anaesthesia. The most frequent are: an epidural haematoma [3-6], a trauma to the intervertebral disc, with leakage of its contents by insertion of the needle [7-9], infections by incorrect sterilization of the surgical field or of the instrumentation used [10] and a chemical arachnoiditis from local anaesthetics [11-14]. In the case analysed in this article, the cause is most likely the last of those mentioned: a hyperbaric bupivacaine arachnoiditis administered at a concentration of 0.5%. This conclusion was reached through an accurate diagnosis of exclusion, which leaves no doubt. This is a rather rare occurrence and thus it was difficult to identify and manage. What follows is an analysis of the clinical path that led to the exclusion of the other causes. The therapeutic measures chosen to treat the patient are also described. Similar complications should not be underestimated, not only for the direct pathological consequences, but also for the psycho-emotional impact on the puerpera. Women in the post-partum are indeed more exposed to depressive manifestations, even serious, of mood [15].

**CASE PRESENTATION**

**Anamnesis**

The clinical case concerns a 34-year-old patient never having given natural birth, at her third pregnancy but with a previous caesarean section and admitted for starting labour at 38+4 weeks of amenorrhoea. The patient, 160 cm tall, weighed 65 kg, 13 of which gained during pregnancy. Her obstetric anamnesis reported a caesarean section in 2010, performed at 38 weeks of gestation for the foetus in the breach position without any prior or posthumous complications, in subarachonoid spinal anaesthesia. The obstetric history reported that there was also a spontaneous abortion in 2018, at 8 weeks of amenorrhoea. On this occasion, clinical investigations and revision surgery of the uterine cavity were performed. A control ultrasound scan did not reveal any alterations to the bladder and sphincter and the uterine-bladder section resulted normal (Figure 1). At the time of admission, the patient denied any hereditary family pathologies and did not present pathologies to organs or apparatuses. Regular intestinal tract activity, physiological diuresis. The patient denied alcohol abuse, drug use or smoking habits. She had never suffered from any neurological, motor or urological problems. The last gestation, narrated in this case report, had a regular course. From the 35th week of gestation, there was only an increase in diastolic pressure up to a maximum of 90 mmHg, though without any need for medical therapy as the haemato-chemical and urinary values were within the limits of the norm.

**Anaesthesia and surgical technique**

On the day of admission, the woman underwent her caesarean section, with a standard procedure and without any complications, for a total duration of 70 minutes. Prior to the surgical procedure a subarachnoid spinal anaesthesia was performed. With the patient seated, and after careful skin disinfection with 10% betadine and a sterile field set-up, local anaesthesia was injected with 1% mepivacaine in the L3-L4 interspace (according to anatomical landmarks), after drying the skin region with a sterile gauze. A subarachnoid block was performed with a 25 G needle: single dural puncture, a leakage of limpid...
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Post-surgery time

On the second day, the woman was passing wind, therefore the urinary catheter was removed, and the intravenous fluid infusion was interrupted and replaced with a semi-liquid diet. After around three hours, the patient showed no signs of urination stimulus, the pelvic region appeared globular and incompressible. A trans-abdominal bladder ultrasound examination was performed, 1100 ml of urine were present: the bladder was over-distended. Temporary catheterisation was performed with a gradual and intermittent emptying to avoid vagal reactions. In the absence of urination stimulus, it was decided to apply a fixed bladder catheter. A two-way silicone Foleya device with a diameter of 14 Charrière was used. The patient began bladder gymnastics with intermittent opening of the catheter and took 25 mg deltacortene tablets (1 tablet/day), as there was the suspicion of possible urethral oedema. The woman also complained of dysesthesia in the buttock and at the back of the left thigh as early as 5 hours after surgery. During the following days, the patient presented constipation. Constipation also arrived over the following days. The urination stimulus remained absent during both bladder gymnastics and in a further attempt to remove the bladder catheter, during which the urinary volume was constantly checked echographically to avoid further over-distension of the bladder. On the fourth day, an additional transvaginal ultrasound was performed, which confirmed a normal uterine involution and the absence of haematomas interposed between the uterus and the bladder. On the fifth post-operative day, a routine post-operative x-ray of the abdomen and pelvis was performed to verify the remote possibility of intestinal obstruction or the presence of foreign bodies (radiopaque), which may induce a mechanical compression or inflammation, although the count of gauzes and instruments on the operating table was correct and had not suggested problems of this kind [16]. The x-ray was negative. On the sixth day after the caesarean section, the symptoms were stable. After attempting a further removal of the bladder catheter to control the resumption of the urination stimulus, a neurological consultation was required. In this specialist visit, the following emerged: absence of central motor deficits (exploration in both Mingazzini and Barré signs) and segmental motor deficits (radicular), with the presence of symmetrically hyperevocable ROTs (Osteo-Tendon reflexes). RCP (Cutaneous plantar reflex) in bilateral flexion. Left Lasegue sign weakly positive after 80° and contralaterally negative. No disturbance in balance or coordination. The picture, according to the neurologist, was attributable to an incomplete inferior cauda equina syndrome (prevalent bladder and sensory involvement in the left S1 region). Abdominal and pelvic magnetic resonance imaging (MRI) was required, given the futility of carrying out an electromyographic study (EMG) with the suspicion of central damage. Lumbosacral and dorsal MRIs were initially performed without a contrast medium.

In MR only the basic T1 and T2 sequences were performed. Nothing significant resulted from this investigation: intervertebral disc normally in place, vertebral canal of normal width and normal representation of the spinal cord and the cauda equina. Thus, the presence of haematomas, spinal infarctions, traumas or abscesses was excluded.

The patient underwent a lumbar MRI with and without contrast using a 1.5T General Electric Medical System. Sagittal FSE-T2 weighted images (TR 3000; TE 114.65), with and without fat-suppression, and sagittal and coronal FSE-T1 weighted images (TR 8.916; TR 340), both before and after contrast administration (gadoteric acid, 11ml Dotarem, 0.2 ml/kg) were employed. This study ruled out the possibility of spinal epidural haematoma, spinal infarction, spinal abscess and disc herniation (Figure 2). Contrast enhanced FSE-T1 weighted images showed intense abnormal enhancement of the nerve root from the T11-T12 level, without either thickening of the nerve roots nor spinal cord abnormalities (Figure 3). During the hospital stay, haematochemical tests were negative for inflammatory processes (negative leukocytes, negative PCR and PCT) and/or for coagulation dyscrasias. The auto-immunity aspect was also investigated by researching ANA, ENA, SSA, SSB, ANTI-ds DNA, GM1, GD1a, GD1b, as suggested by the neurologist. The results of all of these haematochemical tests were negative.
Although the patient presented a minimal bladder sensitivity and a slight urination stimulus desire towards the twelfth day, she was unable to empty her bladder with spontaneous urination. High bladder residues > 450 ml were registered by transabdominal ultrasound (Figure 4).

On the fifteenth day the urodynamic examination was performed. The maximum functional capacity was 246 ml, reduced uroflowmetry values, post urination residue 246 ml in the absence of genital prolapse. In evidence, therefore, a stable cystometrygram, increased bladder sensitivity, reduced capacity and normal compliance. During the patient’s attempts to urinate, no valid contraction of the detrusor muscle was detected. Urination did not occur even with the use of the abdominal press (Figure 5).

The urologist decided to reposition the bladder catheter, scheduling a control after one month. In the absence of any resumption of the urinary flow, the patient would have started intermittent self-catheterization in anticipation of implanting a sacral neuromodulator if the condition persisted for a further 3-4 months. The patient was discharged to home confinement with a bladder catheter until the urological check-up the following month, with martial therapy and with medical therapy of 1 mg of betamethasone tablets (2 tablets/day). Ten days later, after a check-up at our department, post- urination self-catheterization began to resume the stimulus, even with the presence of urination residues > 150 ml. One month after discharge, no significant improvements were noted at the urological examination, and it was decided to continue prescribing intermittent self-catheterizations and to carry out an additional control 30 days later. At this visit, the patient reported that
she needed self-catheterization only for two weeks following the previous visit, progressively moving from using it from twice to once per day. Furthermore, post urination residue volume measurements were never greater than at 150 ml. During the ultrasound there was, after a spontaneous urination of 279 ml, a residue of only 20 cc.

For this reason, it was understood that the patient had a fully recovered bladder function. However, slight skin paraesthesia and slight constipation remained, so she was invited to subsequent checks and a dietary control for the remaining problems of defecation and constipation.

Today, the patient has spontaneously and completely recovered from the urinary and defecating disorders; only slight dysesthesia remains in the posterior region of the left thigh. The bladder recovery required, therefore, about 10 weeks. The alvus returned to being regular after about 14 weeks and the dysesthesia had constant and progressive improvement, with complete resolution only in the gluteal region (after a total period of 22 weeks). The patient has currently no sacral biofeedback device implanted and is taking a supplement with alpha-lipoic acid, acetyl L-carnitine and coenzyme Q10. An MRI control scan was not performed.

**DISCUSSION**

Post-partum urinary retention, also known as puerperal urinary retention, is a condition that is quite frequently brought to the attention of the gynaecologist or midwife. According to Kermans et al. [17], the prevalence of urinary retention after a caesarean section compared to that following vaginal delivery is 3.2% for caesarean and 2.1% for vaginal delivery. Due to scanty data present in the literature, it is not certain whether a caesarean section could be considered a higher risk factor for urinary retention or not. In the study referred to, post-partum urinary retention is defined as the difficulty in spontaneous urination for six hours after vaginal delivery, or six hours after the removal of the Foley catheter after a caesarean section (the catheter is removed 24 hours after surgery), measuring, with ultrasound techniques or with bladder emptying by catheterization, a post urinary residue > 150 ml. On the other hand, it is assumed that the risk associated with a caesarean delivery is higher than that related to natural childbirth for the following reasons:

- the indications leading to the intervention constitute a risk factor for post-partum urinary retention (for example prolonged labour, difficult birth, positive anamnesis for multiple caesarean sections with bladder adhesions to the lower segment of the uterus);
- the incision itself causes oedema and ecchymosis of the bladder in the uterovesical area, moreover there is a prevalence of accidental cystostomy, which varies between 0.14 and 0.31%;
- postoperative immobility, pain caused by the wound and lack of privacy can add to the other risk factors;
- the adverse effects of spinal anaesthesia, due to both its implementation and the drugs used [18].

At the same time, as the state of urinary retention of our patient was detected, neurological symp-
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Symptoms were observed, such as dysesthesia of the left lower limb and ipsilateral buttock and alteration of the rhythm of the alvus, regular until then. These symptoms led us to believe that the pathogenesis could be more at the level of the nervous system than at the visceral level. As a result, it was suspected that the nerve fibres of the cauda equina were involved. The cauda equina syndrome is a condition that can already be diagnosed on a clinical basis, second-level investigations are more than anything used to manage the pathology. It is characterised by five signs and symptoms, not always all present, namely: bilateral neurogenic sciatica, a reduction of perineal sensitivity, alteration of the bladder function, which can reach urinary retention with no perception of urination stimulus, changes in anal tonality and sexual dysfunction.

In a study conducted by Todd and Dickson [19], the cauda equina syndrome (CES) is divided into four groups: • CESC (complete): total loss of cauda equina functionality, absence of perineal sensitivity, loss of anal tone, bladder and rectal paralysis; • CESS (suspected): with bilateral radiculopathy; • CESI (incomplete): with urinary difficulties of neurological origin, loss of urination stimulus, reduced flow and emptying of the bladder with use of the abdominal press; • CESR (with retention): it can be an incomplete evolution in which there is complete deterioration of the nerve fibres, leading to urinary retention devoid of stimuli or a feeling of bladder distension with paradoxical neglect.

According to this classification, we can assert that the patient who came under our observation suffered from incomplete cauda equina syndrome. In our case, it is probably attributable to an anaesthesiologic manoeuvres, the only ones which manipulated the vertebral column and the spinal cord. The possible aetiologies of CESI are: epidural haematoma, rupture of an epidural abscess, infectious causes, direct or indirect trauma from inserting the needle for spinal anaesthesia, chemical arachnoiditis from local anaesthetics, detergents, antiseptics or other substances accidentally introduced in the spinal canal [11]. The hypothesis of chemical arachnoiditis, by exclusion, represents the most probable cause of our clinical case. MRI with gadolinium has actually demonstrated an inflammatory state of the cauda equina. Contrast-enhanced lumbar MRI showed intense abnormal enhancement of the cauda equina.

With the passing of time, scientific literature has been enriched with cases reporting the occurrence of cauda equina syndrome following the administration of bupivacaine, with permanent or, more often, temporary effects [20-29]. It has been shown that spinal anaesthesia does not have significant advantages over general anaesthesia, albeit in the context of laparoscopic gynaecological surgery [30]. In the obstetric field, however, the systemic diffusion of the anaesthetic allows it to reach the foetus through the placenta. For these very reasons, spinal anaesthesia is the preferred choice. In vitro and animal studies confirm the potential neurotoxicity of local anaesthetic drugs [11, 14, 31-36]. The toxicity mechanism seems to be mediated by the release of glutamate in the cephalorachidone fluid, which does not reach significantly different concentrations between the various local anaesthetics, although they have different harmful effects. All studies agree in identifying lidocaine as the most neurotoxic substance; ropivacaine, on the other hand, as the safest [37, 38]. It is estimated that neurotoxicity does not normally occur before the application of twice greater than normal medical use doses of lidocaine and four times greater of tetracaine, bupivacaine and eropivacaine. The detectable histopathological damage to the cauda equina corresponds to axonal degeneration, areas of central necrosis within the spinal cord and sub-pial vacuolization. The extent of these lesions is not related, however, to the functional damage [39].

In another study, bupivacaine produced significant cell death in rats at low concentrations but in the event of prolonged exposure (100 μM or 0.003%). Prolonged extraneural exposure of rat sciatic nerves to bupivacaine also caused significant demyelination and infiltration of nerves with inflammatory cells [40, 41]. This shows that bupivacaine can have harmful effects even at low concentrations in the case of increased exposure time.

In chemical arachnoiditis, the quantity of aspirated cephalorachidone fluid and the spatial distribution of the anaesthetic can also play a determining role in the induction of harmful effects [24].

The use of peripheral perimedullary analgesic techniques is a reality that the gynaecologist is increasingly facing. It is therefore important that they know and recognize any, albeit very rare, side effect of these techniques. The search for the reduction of labour pain and of its duration [42] is a strongly felt need. In the delivery room it is now common to use bupivacaine even for epidural anaesthesia. In 2015 Burgio et al. confirmed that epidural anaesthesia
during labour does not even increase the risk of post-partum urinary incontinence [43]. It is essential to encourage their use in cases where labour is an unpleasant experience (epidural anaesthesia) or in situations where it is necessary to resort to caesarean section (spinal anaesthesia).

In our case, the improvement occurred after about 8-10 weeks. Minimally invasive treatment or intermittent catheterization was sufficient. Adopting this regime of waiting, we avoided neuromodulation neurosurgery, a solution that would have been considered around the sixth month of non-regressive symptoms. Our case teaches that it is important to make patients aware, at the time of informed consent, of the existence of this complication, which is completely independent of the doctor’s intervention and unpredictable, being a purely subjective and random reaction to local anaesthetics causing impaired peri-neural permeability and oedema of nerve fibres. Moreover, careful observation by obstetricians and gynaecologists of post-anaesthesia neurological symptoms is necessary, since they too often focus on mere obstetric-gynaecological controls.

CONCLUSIONS

It is worth underlining that neurotoxicity from local anaesthetics is an unusual, potentially reversible condition: therefore, spinal anaesthesia still proves to be a safe and absolutely practicable method, even if not totally free of serious risks. The real data can differ a lot from the statistical data, which, due to the scarcity of relevant studies, are not particularly significant from a probability viewpoint and in the definition of a prognosis and a standardized therapeutic path to follow. The decisions concerning long-term follow-up are, therefore, placed in the hands of the curing physician. Together with the patient, they will have to decide which is the most suitable position to adopt following the “golden rule”: effectiveness, necessity and least invasiveness possible.

COMPLIANCE WITH ETHICAL STANDARDS

Authors contribution

A.D’A.: study design, clinical participation, supervision of study and manuscript. A.S.: study design, collection of data, clinical participation, writing the manuscript. S.M.: clinical participation, supervision of the manuscript. M.L.: clinical participation, supervision of the manuscript. A.D.S.: clinical participation, supervision of the manuscript. C.D.F.: supervision of the manuscript. M.G.: study design, clinical participation, supervision of study.

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Study registration

N/A.

Disclosure of interests

The authors declare that they have no conflict of interests.

Ethical approval

The manuscript describes a clinical case observed in the San Salvatore Hospital in L’Aquila during the period of hospitalization and follow-up of a patient. No research aim has led to the deviation from clinical management according to scientific evidence. For these reasons, the case report did not require the approval of the Internal Review Board of the University of L’Aquila.

Informed consent

An informed written consent was obtained from the patient before describing the case-report.

Data sharing

Data are available under reasonable request to the corresponding author.

REFERENCES


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