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Management of Two Cases of Spina Bifida and Neonatal Genital Prolapse at the University Hospital of Parakou and Review of the Literature

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Abstract

Spina bifida, or spinal dysraphism, is a malformative pathology related to an anomaly in the development of the nervous system, occurring during embryogenesis. The neural tube does not close properly around the 28th day of life and affects the development of the spinal column and spinal cord. Spina bifida is characterised by damage to the nervous system and will generate handicaps and damage of varying degrees: neurological motor, sensory, cognitive, genito-phincter (bladder and anorectal) deficits with consequences for the quality of life of these people. The literature describes the association between spinal dysraphism and genital prolapse. However, genital prolapse is an exceptional and rare entity in newborns. We report the observations of two newborns: the first case of a newborn born at term, at 7 days of age, who presented a prolapse of the uterine cervix in association with myelomeningocele, without any neuromuscular repercussions, and the second case of a newborn at 10 days of age, presenting with a lumbosacral spina bifida and a uterine prolapse. They benefited from conservative medical treatment characterised by manual reduction of the prolapse in both cases with a favourable evolution. In the case of spina bifida, a cure of myelomeningocele was performed surgically with simple postoperative course.

Keywords

Spina Bifida, Genital Prolapse, Neonate, Complications, CHUD/BA

1. Introduction

Spina bifida, or spinal dysraphism, is a malformative disorder of the nervous system resulting from an abnormality in the development of the nervous system, occurring at an early stage of pregnancy. The neural tube (tubular outline of the central nervous system) does not close properly and affects the development of the spinal column and spinal cord. A neural tube closure defect is characterised by damage to the nervous system and will generate so-called "associated" handicaps, with consequences of varying degrees: neurological motor, sensory, intellectual, bladder and bowel deficits with a significant impact on the quality of life of these people. The exact cause is not known but the involvement of vitamin B9 (or folic acid) taken in sufficient quantities reduces the risk. Certain medications during pregnancy increase the risk of dysraphism.

Genital prolapse is a rare entity in the newborn. An association with congenital malformations of the central nervous system has been reported in the literature. We report the clinical observations of two full term newborns with cervical prolapse and uterine prolapse in association with lumbar myelomeningocele, without neuromuscular repercussions and treated at the Centre Hospital Universitaire et Départemental de Borgou Alibori (CHUD/BA) of Parakou.

OBJECTIVES: To report the clinical and therapeutic particularities of this rare association through these two observations.

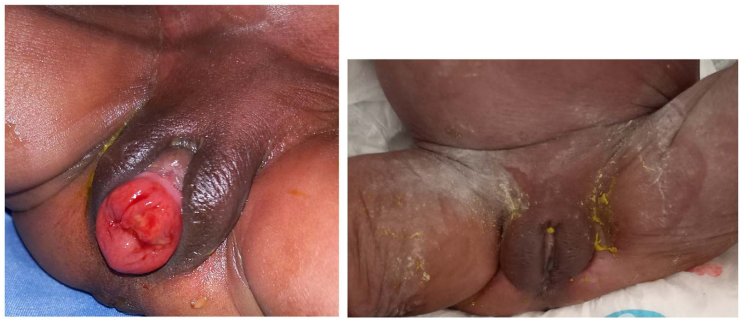
2. Patients and Method

CASE 1

This is a female newborn from a non-consanguineous marriage, in a primiparous, primigravida patient, 22 years old, without any particular pathological history and having carried a pregnancy to term, irregularly monitored. The baby was born by vaginal delivery, with a birth weight of 2420 g, and had an APGAR score of 10/10. She was admitted to our facility on the fourth day of life with a neonatal fever of 39 degrees. The physical examination revealed a dorsolumbar swelling with ulceration of the skin in the centre and a tuft of hair on the cranial part of the swelling. No sensory-motor deficits in the pelvic limbs were observed. Examination of the perineum revealed a prolapse of the uterine cervix through the vaginal orifice (**Figure 1**). The ureteral orifice was normal and the anal sphincter tone was preserved. The clinical malformative work-up concluded that the baby had an incipient macrocrania with bulging anterior and posterior fontanelles indicating incipient hydrocephalus. The archaic reflexes were preserved with an axial tone present. Abdominal and pelvic ultrasound did not reveal any abnormality apart from the absence of visualization of the uterus. In view of the infectious clinical and biological syndrome with a positive cytobacteriological examination of the cerebral spinal fluid, the diagnosis of meningitis caused by staphylococcus was retained and a bi-antibiotic therapy was started. The CT scan of the lumbar spine confirmed spina bifida on 2 levels T12 and L1. In addition to antibiotic therapy, a cure of the myelomeningocele (**Figure 2**) was per-

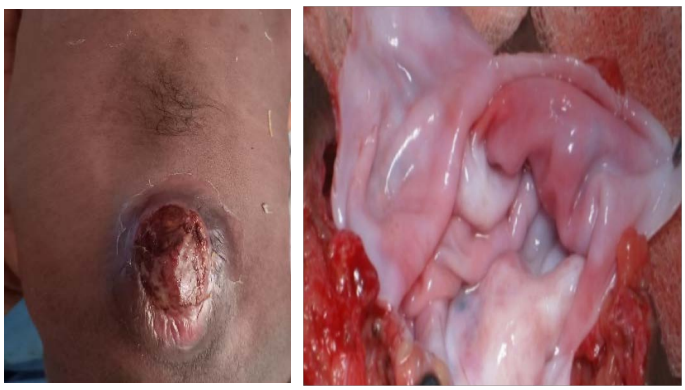
formed 3 weeks later. In emergency, a digital reduction of the prolapse uterin was performed, with placement of a sterile pad for 48 hours. The evolution was favourable without recurrence over a follow-up period of six months.

For the hydrocephalus, he underwent 1 month later to a ventriculo-peritoneal shunt when the cerebral spinal fluid was steril with favourable results.

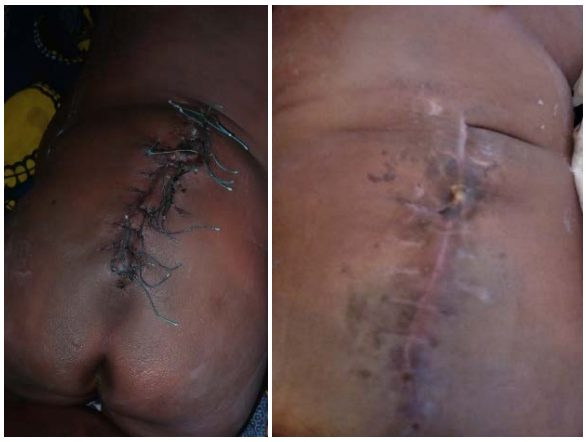


(a) (b)

Figure 1. A genital uterine prolapse (a) and after manual reduction, control after 48 hours (b).



(a) (b)



(c) (d)

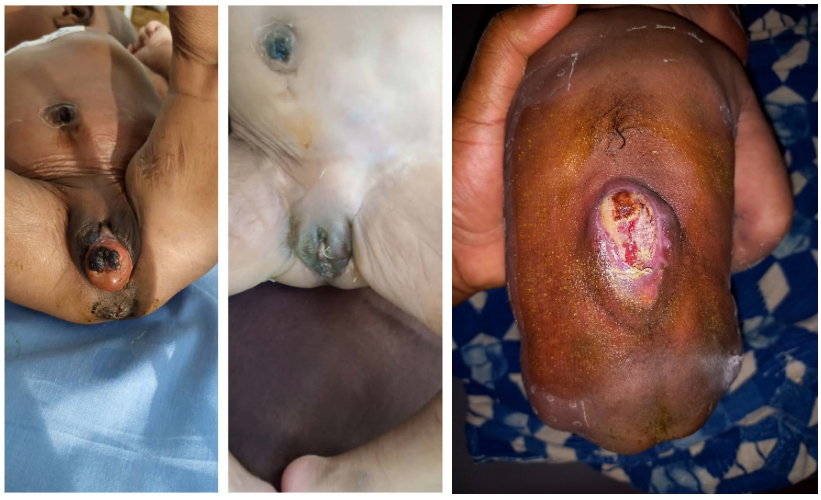
Figure 2. Thoraco lumbar spina bifida (a) with per operative dissection of myelomeningocele (b). Post operative aspect (c). Aesthetic result after 3 months (d).

CASE 2

This is a female newborn, admitted at 2 days of age, born after an ongoing pregnancy, for a genital anomaly associated with spina bifida. The perineal examination found a genital mass suggestive of uterine prolapse with the presence of a crust on the cervix, a sign of mucosal dryness. No associated rectal prolapse (**Figure 3**). The neurological examination did not reveal any motor deficit in the pelvic limbs. The lumbar CT confirmed L1 spina bifida and the brain CT returned normal. Additional CT scans of the abdomen did not reveal any intra-abdominal malformations. She underwent emergency manual reduction of the genital prolapse after moistening the cervix and disinfection to remove the crust. A sterile tampon was kept in place for 48 hours. The evolution was favorable, without recurrence of the genital prolapse. She was operated on for her spina bifida with a favourable evolution and a satisfactory aesthetic result.

3. Discussion

Spina bifida is a congenital malformation with incomplete closure of the neural tube (at the end of the first month of embryogenesis) followed by incomplete closure of the last vertebrae. The frequency is currently 1/2000 births in France. The highest frequency is found in English-speaking countries (up to 6/1000). The favourable factors are now known are: genetic (PAX 3 gene) [1]; deficiency (lack of vitamin B9 and zinc during the first month of pregnancy); metabolic (insulin-dependent maternal diabetes); thermal (fever and baths that are too hot during the first month of pregnancy). Antenatal detection by ultrasound is only 69% reliable. Significant progress is expected with three-dimensional images. The consequences vary in severity. Some of them are underlying the malformation: sensory-motor paralysis of the lower limbs (from S2 to D12); orthopaedic deformities (talus, knee flessum, hip dysplasia with a high risk of dislocation for



(a) (b) (c)

Figure 3. (a) Uterine prolapse with dryness crust. (b) Image after manual reduction. (c) Thoraco lumbar spina bifida.

the L4 level, hyperlordosis, scoliosis) [1] [2]; osteoporosis with a risk of spontaneous fracture of the lower limbs; bladder and bowel disorders (incontinence and retention); genito-sexual disorders (especially in men). Others are overlying the malformation: hydrocephalus (often requiring early shunting) [3] [4]; Arnold-Chiari type II malformation (often affecting higher functions and vision). The others are of a general nature: multiple allergies (particularly to latex); endocrine disorders (ectopic testicle, advanced puberty in girls); overweight. In the two cases reported, the newborns presented genital prolapses with the first child developing early hydrocephalus and an infectious complication requiring appropriate antibiotic therapy; for the hydrocephalus, she benefited from a ventriculoperitoneal shunt and a cure for myelomeningocele [5].

The management of these "congenital hydrocephalic paraplegics" is complex, multidisciplinary and long-term [5] [6] [7]. There is a need for regular follow-up in neurosurgery [5], orthopaedics and urology, and for rehabilitation sessions [1] [8]. We insist on the quality of the management of vesico-renal problems, whatever the motor level [9] [10] [11], because they condition the functional and then vital prognosis of these patients in the long term. For the second newborn, after manual reduction of the prolapse and surgical cure of her myelomeningocele, the outcome was favourable without recurrence of the prolapse.

4. Conclusion

Newborn uterine prolapse is an exceptional complication associated with spina bifida. The treatment is manual reduction of the prolapse structure with good outcome, followed by myelomeningocele surgery. Acid folic taking during the period before and the first month of pregnancy must be an important recommendation for spina bifida prevention.

Ethical Aspect

Parental consent has obtained for the use of patient data, and no image allows identification.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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