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Original article

Neurodevelopmental long-term outcome in children with hydrocephalus requiring neonatal surgical treatment

Développement psychomoteur à long terme des enfants opérés d'une hydrocéphalie néonatale

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ABSTRACT

Purpose. – To assess long-term neurodevelopmental outcome in children with hydrocephalus requiring neurosurgical treatment during the neonatal period.
Methods. – This prospective longitudinal population-based study included 43 children with neonatal shunted hydrocephalus. The 43 children were prospectively reviewed in the presence of their parents at the outpatient clinic. Cognitive and motor outcomes were assessed respectively using different Wechsler scales according to age and Gross Motor Function Classification System (GMFCS). Postoperative MRI was routinely performed.
Results. – The mean gestational age at birth of the 43 consecutive children with neonatal hydrocephalus (sex ratio M/F: 1.39) was 34.5 ± 5.4 weeks of gestation. At mean follow-up of 10.4 ± 4 years, mean total IQ was 73 ± 27.7, with equivalent results in mean verbal and mean performance IQ. Of the 33 children with IQ evaluation, 18 presented an IQ ≥ 85 (41.9%). Efficiency in walking without a mobility device (GMFCS ≤ 2) was obtained in 37 children (86%). Only severity of postoperative ventricular dilation was significantly associated with unfavorable outcome (Evans index > 0.37; odds ratio: 0.16, $P = 0.03$).
Conclusion. – This information could be provided to those families concerned who often experience anxiety when multi-disciplinary management of neonatal hydrocephalus is required.

Mots clés :
Hydrocéphalie
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Nouveau-né
Dérivation ventriculo-péritonéale
Ventriculocysternostomie
Déficit cognitif
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Performances intellectuelles

RÉSUMÉ

Objectif. – Évaluer les résultats à long terme du développement psychomoteur des enfants avec une hydrocéphalie néonatale opérée.
Patient et méthode. – Cette étude prospective longitudinale incluait 43 enfants opérés dans le cadre d'une hydrocéphalie néonatale. Ces enfants ont été évalués de manière prospective en présence de leurs parents. Les performances cognitives et motrices étaient évaluées respectivement par les échelles de Wechsler et Gross Motor Function Classification System (GMFCS). Des IRM postopératoires étaient réalisées.
Résultats. – L'âge gestationnel moyen de notre population (sex-ratio M/F : 1,39) à la naissance était de 34,5 ± 5,4 semaines de gestation. Après un suivi moyen de 10,4 ± 4 ans, le QI moyen était de 73 ± 27,7, avec des résultats équivalents des performances verbales moyennes et du QI moyen. Des 33 enfants

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évalués sur le QI, 18 présentaient un QI ≥ 85 (41,9 %). La marche sans aide (GMFCS ≤ 2) était possible chez 37 enfants (86 %). Seule la dilatation ventriculaire postopératoire était associée avec un résultat clinique défavorable (index d'Evans > 0,37 ; *odds ratio* : 0,16 ; $p = 0{,}03$).
Conclusion. – Ces données peuvent servir d'aide à l'information des familles de ces patients pour lesquels une prise en charge multidisciplinaire est nécessaire.

1. Introduction

Neonatal hydrocephalus is a life-threatening condition resulting from active distension of the ventricular cavities. It occurs in 2.5–8.2/10 000 live births [1], during the first 28 days of life [2,3]. The predominant etiology is intraventricular hemorrhage (IVH) causing approximately half of all cases of neonatal hydrocephalus followed by congenital malformation (congenital aqueduct stenosis, Dandy Walker syndrome, polymalformative syndrome and spinal dysraphism), meningitis, and rarely tumors [4]. Neonatal hydrocephalus is mainly treated by noninvasive diuretic therapy with administration of acetazolamide and furosemide, by serial subtractive lumbar puncture and if unsuccessful by different invasive procedures of CSF drainage [5–8]. Neurodevelopmental outcome at 2 years of neonatal hydrocephalus requiring invasive procedures was altered in approximately 47% of infants [9–11]. Long-term outcome has been scarcely evaluated to date and remains unclear.

The aim of this study was to prospectively assess long-term outcome in children with hydrocephalus requiring invasive treatment during the neonatal period.

The primary objective was to evaluate the development of intellectual and motor performance in children older than 4 years. The secondary objectives were to assess procedure-related complications, describe postoperative MRI, and determine predictive factors of unfavorable outcome.

2. Patients and method

2.1. Study design

This prospective longitudinal single-center study included all consecutive children who had undergone surgical treatment for hydrocephalus during the neonatal period between January 1993 and December 2007.

2.2. Population

Inclusion criteria were:

- symptomatic hydrocephalus;
- confirmed diagnosis before 28 days of life requiring surgical treatment: ventriculo-peritoneal (VP) shunt or endoscopic third ventriculocisternostomy (ETV);
- children older than 4 years at the time of cognitive evaluation.

Exclusion criteria were:

- diagnosis of symptomatic hydrocephalus after 28 days of life;
- ventricular enlargement without any evidence of hydrocephalus;
- parental refusal to participate in long-term follow-up.

Over a 15-year period, 53 consecutive children were eligible and 10 were excluded due to death in three, lost to follow-up in five, and parental refusal in two children. This population-based study included 43 children. The institutional Ethics Committee approved the study (Fig. 1).

2.3. Neonatal hydrocephalus management

Diagnosis of neonatal hydrocephalus was suspected in newborns with a large head (more than 2 standard deviations above the mean or greater than 95th percentile on the standard growth curve), bulging anterior fontanel, suture separation, and axial hypotonia [12]. Confirmed by cranial ultrasonography (cUS), the diagnosis of ventricular dilatation was established by recording lateral ventricular ratio (LVR), superior to a value of 0.34 [13]. Routine magnetic resonance imaging (MRI) was used to diagnose causes and associated abnormalities; this examination was performed during the 24 h of the diagnosis without impact on the delay of surgical procedure. Etiology was classified as IVH, central nervous system (CNS) malformations (congenital aqueduct stenosis, Dandy Walker syndrome, spina bifida and polymalformative syndrome), CNS infectious process (meningitis, ventriculitis) or neoplasm.

Surgical treatment was proposed in cases of developing hydrocephalus i.e. increasing head circumference, neurological deterioration, and increasing LVR on successive cUS. Ventricular drainage was routinely carried out using a VP shunt. ETV was preferred for congenital aqueduct stenosis. All patients were monitored pre- and postoperatively in neonatal intensive care unit. Surgical details (timing of drain implantation or ETV, as well as number and indication of revisions) were described and operative complications related to procedures requiring surgical revision were reported.

2.4. Clinical outcome

During 2011, the 43 children in the presence of their parents were prospectively reviewed by a neuropediatrician (CVH, SM) in the outpatient clinic.

Cognitive assessment was evaluated by calculation of Intelligence Quotient (IQ) using different Wechsler scales according to age: Wechsler Preschool and Primary Scale of Intelligence (WPPSI-3) for children aged from 4 to 6 years, Wechsler Intelligence Scale for Children (WISC-4) for children aged from 6 to 16 years, and Wechsler Adult Intelligence Scale (WAIS-4) for children older than 16 years [14]. When Wechsler scales were not applicable for children with high cognitive or intentional deficiency, IQ was considered as an unfavorable cognitive outcome with a minimal IQ score of 40. Cognitive outcome was categorized as favorable for children with an IQ score higher than or equal to 85 and unfavorable for IQ lower than 85. Motor outcome was assessed by the Gross Motor Function Classification System (GMFCS) [15]. We classified motor outcome as favorable for children with level 1 GMFCS (without walking limitation) at the end of the study and unfavorable for the others. Moreover, to detect fine motor function disorder (accuracy, dexterity and visual-manual coordination), the patients were asked to draw a line between two lines and to copy a simple figure. Type of schooling was classified into 3 groups:

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