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Late-onset myelopathy following Torkildsen shunt placement for a midbrain cystic tumor: a case report

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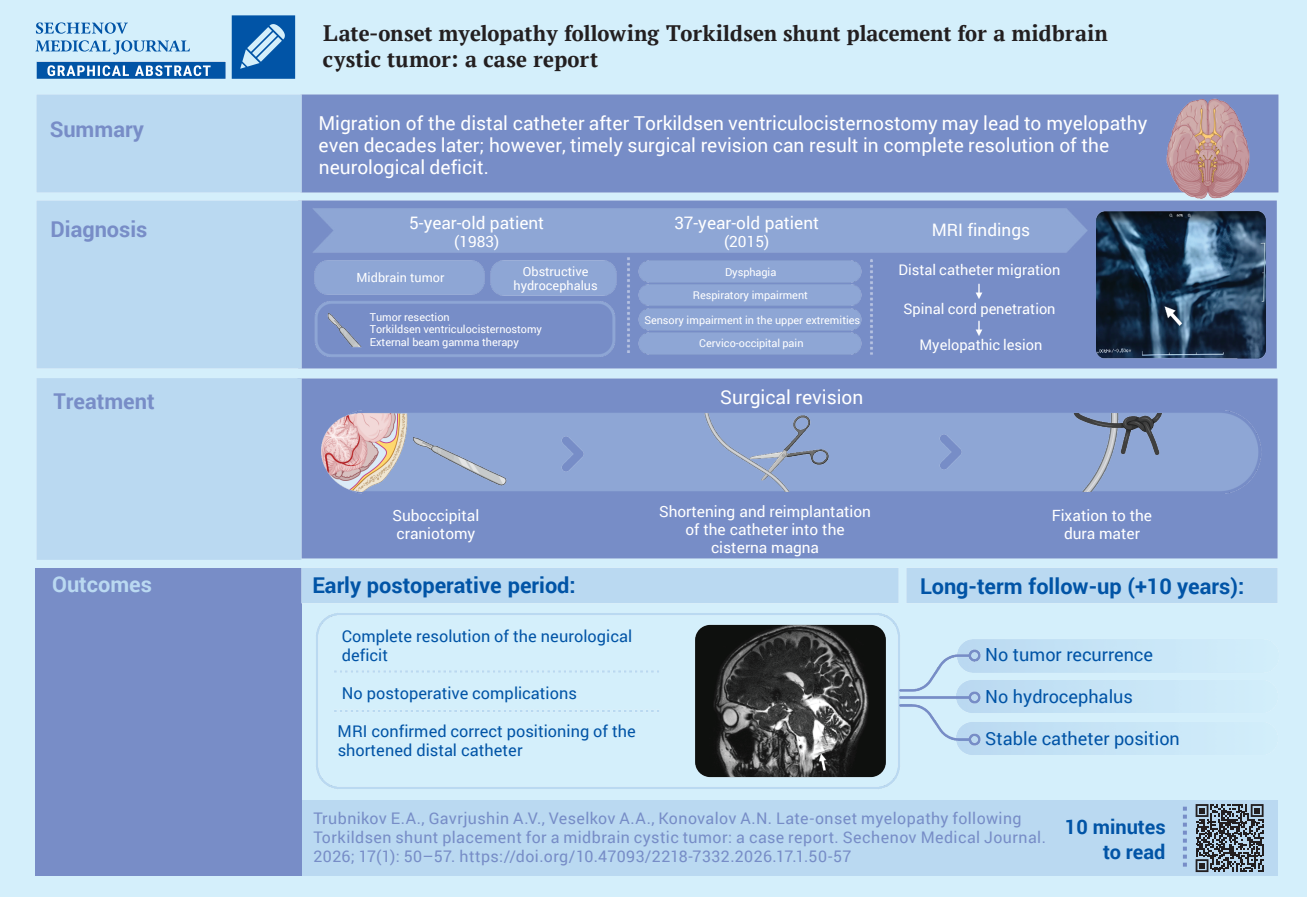

Abstract

Torkildsen ventriculocisternostomy was historically one of the principal surgical treatments for obstructive hydrocephalus. However, nowadays it tends to be regarded mainly as a salvage procedure when standard shunting or endoscopic ventriculostomy is not feasible.

Case report. A 5-year-old boy with obstructive hydrocephalus secondary to a cystic midbrain tumor underwent tumor resection combined with Torkildsen ventriculocisternostomy. Postoperatively, adjuvant radiotherapy was administered, resulting in long-term disease stabilization. At the age of 37 years, 32–33 years after surgery, he developed dysphagia, respiratory disturbances, cervico-occipital pain, and sensory impairment in the upper limbs. Magnetic resonance imaging demonstrated migration of the distal catheter tip with penetration into the upper cervical spinal cord segments and formation of a focal myelopathic lesion. A suboccipital craniotomy was performed; the migrated catheter segment was removed, the system was shortened, and the distal end was reimplanted into the cisterna magna with fixation to the dura mater. Complete regression of neurological deficits was achieved, with a favorable 10-year follow-up.

Discussion. We report a rare delayed case of myelopathy caused by migration of the cisternal catheter tip more than 30 years after Torkildsen ventriculocisternostomy. This observation highlights the need for lifelong surveillance of patients who have undergone such procedures, strict adherence to surgical technique (appropriate catheter length selection and secure fixation), and timely surgical revision at the earliest signs of brainstem dysfunction or involvement of the upper cervical spinal cord.

Keywords: brainstem tumors; occlusive hydrocephalus; Torkildsen ventriculocisternostomy; reimplantation; long-term follow-up
MeSH terms:
CASE REPORTS
BRAIN STEM NEOPLASMS – COMPLICATIONS
BRAIN STEM NEOPLASMS – SURGERY
HYDROCEPHALUS – SURGERY
HYDROCEPHALUS – ETIOLOGY
NEUROSURGICAL PROCEDURES
POSTOPERATIVE COMPLICATIONS – DIAGNOSIS
SPINAL CORD DISEASES – DIAGNOSIS
SPINAL CORD DISEASES – ETIOLOGY
FOLLOW-UP STUDIES
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Миелопатия как отдаленное осложнение операции Торкильдсена у больного с кистозной опухолью среднего мозга: клиническое наблюдение

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Аннотация

Вентрикулоцистерностомия по Торкильдсену исторически была одним из основных методов лечения окклюзионной гидроцефалии, но сейчас рассматривается преимущественно как резервная процедура при ограниченных возможностях для стандартного шунтирования или эндоскопической вентрикулостомии.

Описание случая. У мальчика 5 лет с окклюзионной гидроцефалией на фоне кистозной опухоли среднего мозга выполнено ее удаление с дополнением вентрикулоцистерностомией по Торкильдсену. После операции проведена дистанционная гамма-терапия, достигнута длительная стабилизация. Через 32–33 года, в возрасте 37 лет, возникли дисфагия, дыхательные нарушения, боли в шейно-затылочной области и расстройства чувствительности в руках. Магнитно-резонансная томография выявила миграцию дистального конца катетера с инвазией в верхние шейные сегменты спинного мозга и формированием очага миелопатии. Выполнена субокципитальная краниотомия, мигрировавший сегмент катетера удален, система укорочена, дистальный конец реимплантирован в большую затылочную цистерну с фиксацией к твердой мозговой оболочке. Отмечен полный регресс дефицита и благоприятный 10-летний катамнез.

Обсуждение. Представлен редкий отсроченный случай миелопатии, обусловленной миграцией цистернального конца катетера более чем через 30 лет после вентрикулоцистерностомии по Торкильдсену. Наблюдение подчеркивает необходимость пожизненного мониторинга пациентов с такими вмешательствами, строгого соблюдения техники (подбор длины катетера, надежная фиксация) и своевременной хирургической коррекции при первых признаках стволовой симптоматики и поражения верхних шейных сегментов спинного мозга.

Ключевые слова: опухоли ствола головного мозга; окклюзионная гидроцефалия; вентрикулоцистерностомия по Торкильдсену; реимплантация; катамнестическое наблюдение

Рубрики MeSH:
ОПИСАНИЕ СЛУЧАЕВ
СТВОЛА МОЗГА НОВООБРАЗОВАНИЯ – ОСЛОЖНЕНИЯ
СТВОЛА МОЗГА НОВООБРАЗОВАНИЯ – ХИРУРГИЯ
ГИДРОЦЕФАЛИЯ – ХИРУРГИЯ
ГИДРОЦЕФАЛИЯ – ЭТИОЛОГИЯ
НЕЙРОХИРУРГИЧЕСКИЕ МЕТОДЫ
ПОСЛЕОПЕРАЦИОННЫЕ ОСЛОЖНЕНИЯ – ДИАГНОСТИКА
МОЗГА СПИННОГО БОЛЕЗНИ – ДИАГНОСТИКА
МОЗГА СПИННОГО БОЛЕЗНИ – ЭТИОЛОГИЯ
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Abbreviations:
MRI – magnetic resonance imaging

HIGHLIGHTS

Early revision surgery with ventricular catheter reimplantation may result in complete recovery from neurological deficits caused by myelopathy secondary to catheter migration.

This case supports lifelong neurosurgical follow-up after Torkildsen ventriculocisternostomy because delayed complications may occur decades later.

These findings support a more selective and judicious approach to the use of the Torkildsen procedure. In contemporary clinical practice, ventriculocisternostomy appears justified only in patients with absolute indications.

Torkildsen ventriculocisternostomy was historically one of the earliest and most widely used surgical procedures for the treatment of obstructive hydrocephalus. However, in contemporary neurosurgical practice it is regarded mainly as a rescue procedure [1]. Nevertheless, patients with functioning or modified Torkildsen shunt systems continue to be encountered in clinical practice, which underscores the importance of assessing long-term complications of these interventions [2].

Clinically significant late complications include migration of the distal catheter tip with penetration into surrounding structures, including the cervical spinal cord, resulting in myelopathy. Such cases are considered extremely rare but potentially disabling and often require revision surgery at the craniovertebral junction [3].

Despite various technical modifications and catheter fixation techniques, the risk of distal catheter dislocation and penetration into adjacent tissues cannot be completely eliminated [4]. In this context, isolated clinical observations with a very long interval after the primary operation are of particular interest for understanding the pathogenesis of these complications, refining revision strategies, and assessing long-term prognosis.

The aim of this report is to present a rare case of distal ventricular catheter migration into the upper cervical spinal cord occurring 33 years after surgery for a cystic midbrain tumor, and to analyze the results of catheter reimplantation together with long-term (10-year) follow-up outcomes.

CASE REPORT

Hydrocephalus first manifested in a 5-year-old boy as diplopia caused by alternating strabismus. His general somatic condition at admission was as expected for his age. A neurological examination revealed brainstem signs, including fine horizontal nystagmus on extreme gaze and static ataxia. Computed tomography demonstrated a midbrain mass with evidence of aqueductal obstruction at the level of the cerebral aqueduct.

The patient underwent surgery. Intraoperatively, a lesion with a predominantly cystic component was identified and resected. The limited amount of biopsy material did not permit histological verification of the lesion. To prevent recurrent hydrocephalus in the event of possible tumor regrowth, the procedure was supplemented with Torkildsen ventriculocisternostomy. The ventricular catheter length was selected according to the patient's anthropometric characteristics. The catheter was secured to the dura mater with interrupted sutures at two points to ensure stable positioning.

Following treatment, the patient's condition was satisfactory. Neurological examination demonstrated persistent mild brainstem signs at the level of the pontine tegmentum, including diplopia and fine horizontal nystagmus. In the postoperative period, a course of adjuvant external beam gamma therapy was administered (20 sessions), with a total focal dose of 40 Gy delivered to the operative field. During subsequent follow-up, the patient remained clinically stable, with no evidence of deterioration.

Clinical deterioration occurred at the age of 37 years. In September 2015, the patient presented with dysphagia, respiratory disturbances, and sensory loss in the upper extremities. These symptoms had been present for one month before admission and had progressively worsened.

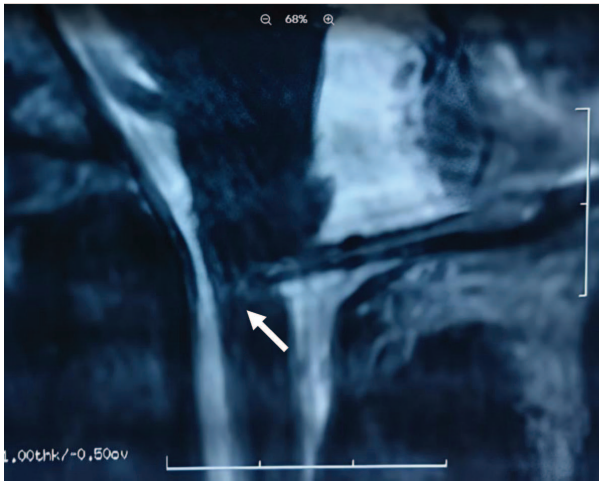
General condition on admission was satisfactory. The patient was alert and oriented, with a Glasgow Coma Scale score of 15; respiratory rate was 17 breaths per minute, heart rate 75 beats per minute, and blood pressure 125/80 mmHg. Neurological examination, in addition to persistent brainstem signs (horizontal nystagmus), revealed impaired proprioceptive and kinesthetic sensation in the upper extremities, accompanied by localized pain in the cervico-occipital region.

Magnetic resonance imaging (MRI) showed no evidence of tumor recurrence but demonstrated migration of the distal ventricular catheter tip with penetration into the spinal cord at the spinomedullary junction (Fig. 1A, B).

The development of neurological deficits associated with migration of the distal catheter tip into the upper cervical spinal cord and the presence of myelopathic lesions on MRI constituted an indication for revision surgery.

The operation was performed on September 30, 2015. A midline suboccipital craniotomy with bone flap replacement was carried out. In the region of the

A



B

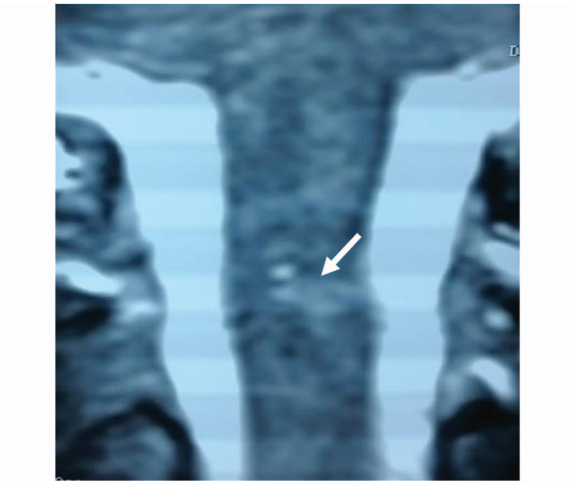
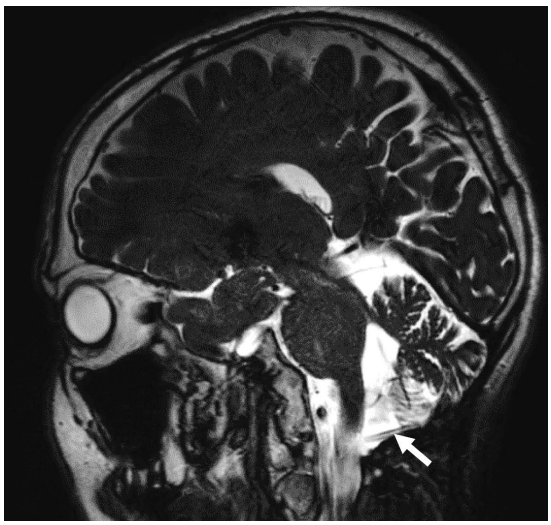


FIG. 1. Preoperative magnetic resonance imaging data of a 37-year-old patient with dislocation of the distal ventricular catheter tip (September 23, 2015).
A. T2-weighted sagittal image demonstrating penetration of the distal shunt catheter tip into the spinal cord at the level of the upper cervical segments (arrow).
B. T2-FLAIR coronal image demonstrating a hyperintense lesion with ill-defined margins corresponding to focal myelopathy (arrow).

A



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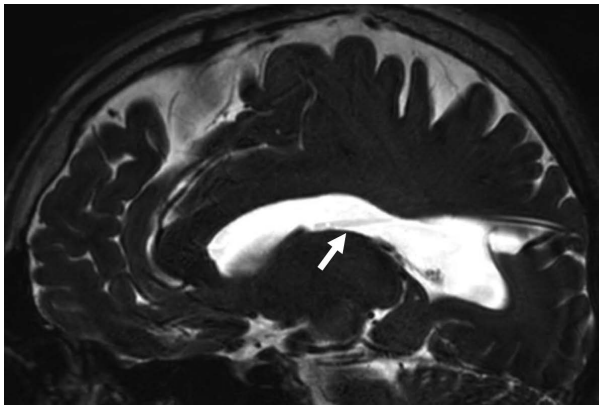

FIG. 2. Magnetic resonance imaging of the brain performed 1 month after surgery (October 29, 2015).
A. T2-weighted sagittal image showing the shortened distal ventricular catheter tip reimplanted into the cisterna magna (arrow).
B. T2-weighted sagittal image showing the proximal ventricular catheter segment in correct position (arrow); the middle third of the catheter courses through the occipital subgaleal space. No evidence of tumor recurrence is seen.

posterior median sulcus of the upper cervical spinal cord, the migrated catheter tip was identified embedded within the spinal cord parenchyma. After removal, the distal catheter segment was shortened and reimplanted into the cisterna magna; the distal end of the ventricular catheter was additionally secured to the dura mater. The postoperative course was uneventful, and the patient was discharged in satisfactory condition on postoperative day 5. At discharge, complete resolution of the neurological deficit was observed, with no new focal neurological signs.

One month after surgery, follow-up MRI confirmed correct positioning of the shortened distal ventricular catheter tip within the cisterna magna, with no evidence of tumor recurrence (Fig. 2A, B).

At the 10-year follow-up after ventricular catheter reimplantation, the patient reported only occasional cervico-occipital pain, with no focal neurological deficits. Annual follow-up MRI demonstrated no significant interval changes: there was no evidence of tumor progression or hydrocephalus, and the ventricular catheter remained in proper position.

DISCUSSION

In the present case, the total follow-up duration was 43 years, representing a rare and virtually unique experience in the management of a patient after Torkildsen ventriculocisternostomy. This highlights the fact that the long-term consequences of this procedure remain clinically relevant because despite the emergence of alternative extra- and intracranial shunting techniques, ventriculocisternostomy and its modifications continue to be used, albeit rarely, in modern neurosurgery [2, 5].

During the first decades of Torkildsen ventriculocisternostomy use, a pooled analysis of 136 published cases by J.E. Scarff [6] showed that arrest of hydrocephalus progression over a two-year period was achieved in 58% of patients, with an operative mortality of approximately 30%. According to N. Morota et al. [7], these unfavorable outcomes in historical series largely reflect the level of neuroimaging, anesthetic, and neurosurgical care available in the mid-20th century, when the procedure was performed as a palliative intervention in severely ill, often terminal patients with advanced obstructive hydrocephalus. Nevertheless, the long-term effectiveness of the technique is supported by isolated case reports with follow-up periods of up to 30 and 50 years, demonstrating durable shunt function and satisfactory neurological outcomes [1, 8].

Complications of Torkildsen ventriculocisternostomy include infectious and nonspecific surgical complications (wound infection, meningitis), mechanical shunt dysfunction (migration, kinking, axial rotation, and catheter obstruction), as well as neurological complications such as myelopathy, tetraparesis, and facial pain caused by penetration of the cisternal catheter tip into the brainstem or upper cervical spinal cord [3, 7, 9].

Myelopathy is considered one of the most serious, although rare, long-term complications of Torkildsen ventriculocisternostomy. It results from migration or progressive intraparenchymal advancement of the distal catheter tip, causing compression of the cervical spinal cord and craniocervical junction. Such cases have been reported both 8–15 years after surgery and as late as 25–30 years or more thereafter. Clinically, patients may present with progressive spastic tetraparesis, sensory disturbances, and signs of medullary involvement, whereas MRI can demonstrate the catheter penetrating the brainstem parenchyma or upper cervical spinal cord. Treatment usually consists of catheter removal, shortening, or repositioning with fixation to the dura mater; however, because of prolonged compression, neurological deficits often resolve only partially, underscoring the importance of early diagnosis and prevention of catheter migration [3, 4, 10, 11].

In the present case, myelopathy developed 32 years after Torkildsen ventriculocisternostomy and was most likely related to inappropriate selection of ventricular catheter length and insufficient fixation to the dura mater, resulting in gradual distal catheter migration with penetration into the brainstem. Surgical correction consisted of revision of the shunt system, removal of the migrated catheter segment, shortening and reimplantation of the cisternal end, and additional fixation to the dura mater.

To prevent myelopathy caused by migration or excessive advancement of the cisternal catheter tip, G. Ehni et al. [3] emphasized the need for careful selection of catheter length according to the patient's age, so that the catheter lies closely along the bone and enters the subarachnoid space of the cisterna magna only minimally. They additionally recommended positioning the distal tip strictly in the midline and securing it firmly to the dura and arachnoid mater with non-absorbable sutures, splitting the catheter tip into two thin "arms" to improve fixation. N. Morota et al. [7], based on their experience in pediatric patients, also highlighted the importance of thorough preoperative MRI assessment of the craniovertebral junction, partial resection of the posterior arch of the first cervical vertebra when necessary, and careful insertion of the catheter through a midline dural opening while limiting its intradural length to approximately 2 cm, thereby reducing the risk of compression of the brainstem and upper cervical spinal cord.

In the present case, the most likely cause of myelopathy was insufficient fixation of the ventricular catheter to the dura mater, which contributed to gradual distal catheter displacement with penetration into the brainstem. Timely diagnosis and surgical correction resulted in rapid and complete resolution of the neurological deficit.

Thus, the role of Torkildsen ventriculocisternostomy in contemporary neurosurgical practice remains debatable in the era of widespread use of endoscopic third ventriculostomy and various ventriculoperitoneal shunting techniques. In most patients, these methods are considered first-line treatment options, whereas the Torkildsen procedure and its modifications may serve as a salvage option in carefully selected cases of obstructive hydrocephalus where anatomical constraints or prior surgery limit the use of standard techniques. In such rare situations, strict adherence to technical principles – including optimal catheter length selection, secure fixation, and thoughtful revision of previously implanted shunt systems – may reduce the risk of severe complications and improve long-term clinical outcomes.

CONCLUSION

The present case demonstrates that Torkildsen ventriculocisternostomy may provide durable long-term control of obstructive hydrocephalus. However, sustained shunt function is associated with a risk of severe delayed complications related to the position of the cisternal catheter. The occurrence of myelopathy decades after surgery underscores the need for lifelong surveillance of patients who have undergone this type of cerebrospinal fluid shunting procedure, as well as timely system revision at the earliest signs of brainstem or cervical spinal cord involvement. Taken together, the available literature and the present case suggest that, in contemporary neurosurgery, the Torkildsen procedure should be regarded primarily as a reserve option for a limited subset of patients with obstructive hydrocephalus, whereas endoscopic third ventriculostomy and ventriculoperitoneal shunting remain the standard first-line treatments. In the rare situations in which ventriculocisternostomy is selected, meticulous surgical technique–including precise catheter length selection and secure fixation–is essential to reduce the risk of late complications and achieve favorable long-term outcomes.

AUTHOR CONTRIBUTIONS

Alexander N. Konovalov developed the concept. Elisey A. Trubnikov prepared the draft of the manuscript. Andrey V. Gavrjushin and Aleksei A. Veselkov contributed to the reviewing and editing of the text. Aleksei A. Veselkov and Elisey A. Trubnikov participated in data collection and patient examination. Andrey V. Gavrjushin performed the formal analysis. Alexander N. Konovalov was responsible for the methodology and provided scientific supervision throughout the study. All authors have approved the final version of the article.

ВКЛАД АВТОРОВ

А.Н. Коновалов разработал концепцию рукописи. Е.А. Трубников подготовил черновик рукописи. А.В. Гаврюшин и А.А. Веселков внесли вклад в рецензирование и редактирование текста. А.А. Веселков и Е.А. Трубников участвовали в сборе данных и обследовании пациента. А.В. Гаврюшин выполнил формальный анализ. А.Н. Коновалов отвечал за методологию и осуществлял научное руководство на протяжении всего исследования. Все авторы одобрили окончательную версию статьи.

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