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Good results of neurorehabilitation of ischaemic stroke — locked-in syndrome in a pregnant woman with delivery of healthy child

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To the Editors

The mortality rate in locked-in syndrome (LIS) is significant. Patients usually die after a few days, and up to 87% of deaths occur within the first four months. If patients survive the first year, 86% of them will still be alive four years later. LIS patients have low quality of life scores, mainly due to motor dysfunction [1].

A 21-year-old woman in the 8th week of pregnancy was admitted to the Accident & Emergency Department (A&E) of her local District Hospital (DH) due to headache accompanied by dizziness, vertigo, balance disorders, and numbness of the upper and lower limbs that had been increasing gradually for c.16 hours. Speech disorders and limb ataxia were observed, and verbal contact with the patient had become difficult. Before the incident, the patient had been healthy, without addictions, and was not taking any medications. CT scan and CT angiogram of the head showed a stroke in the left cerebellar hemisphere (Fig. 1A, B) and a thrombus in the basilar artery (Fig. 1C).

Initially admitted to the Department of Neurology of the DH, due to increasing disturbances of consciousness and breathing, the woman was transferred to the Intensive Care Unit (ICU) of the University Medical Centre (Uniwersyteckie Centrum Kliniczne, UCK) in Katowice, Poland c.12 hours after arriving at A&E.

In the ICU, the patient was unconscious, with respiratory and circulatory failure. A divergent position of the right eyeball, vertical nystagmus, flexion reaction to pain in the right limbs, extension in the left limbs, and bilateral Babinski sign were observed (NIHSS = 33 points, mRS = 5 points, GCS score = 6 points). Due to the exceeded time window (i.e. more than 24 hours) and the presence of an ischaemic area visible in CT and CT angiography, the patient was not qualified for either thrombolysis or thrombectomy according to the guidelines of the Polish Neurological Society [2]. In the ICU, low molecular weight heparin (LMWH) 5,000 U.I./day and ASA 150 mg/day were administered. After 20 days, these doses were reduced to LMWH 2,500 U.I./day and ASA 75 mg/day. The patient

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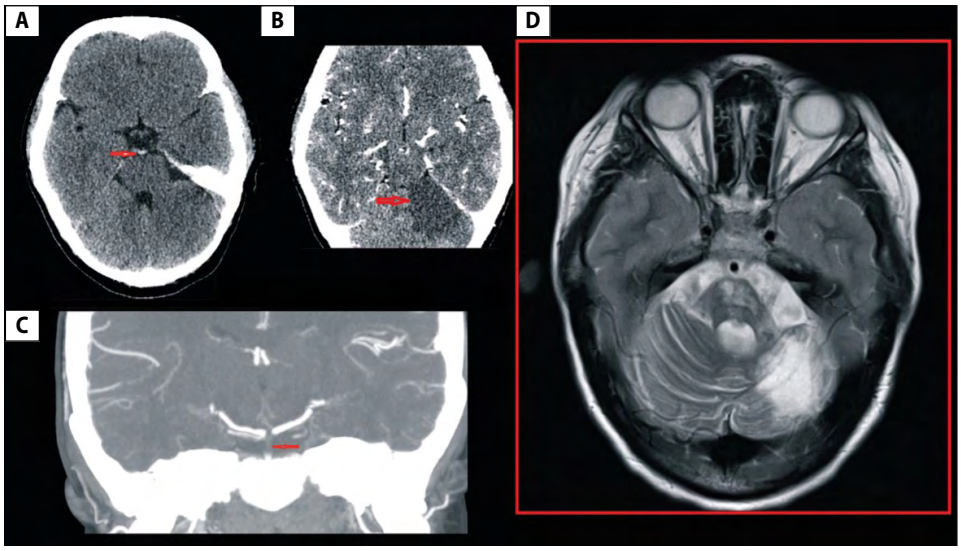

Figure 1. Neuroimaging examinations performed at Accident & Emergency Department of a district local hospital, and an ambulatory follow up two years after the onset of symptoms. **A.** Head Computed Tomography (CT) scan: hyperdensity of the basilar artery and an early signs of the left cerebellar hemisphere ischemic stroke; **B.** Head Angio-CT scan: irreversible left cerebellar hemisphere damage – ischemic stroke; **C.** Head Angio-CT scan: 9 mm in long axis thrombus in the basilar artery. Posterior cerebral arteries were supplied by posterior communicating arteries; **D.** Brain MRI (2 years after the stroke): malacic lesions in the upper part of the left cerebellar hemisphere, in middle cerebellar peduncle, in the central part of the pons and partially in the medulla *(courtesy of Helimed-Diagnostic imaging)*

Table 1. Specific laboratory test results

Test		Result	Unit	Reference range
Activity of S-protein		66	%	64–126
Activity of C-protein		> 149.9	%	70–140
Homocysteine		5.36	µmol/L	4.44–13.56
Activity of factor VIII		212.1	%	70–150
Activity of factor IX		130.3	%	70–120
Anti-cardiolipin antibodies	IgG	< 2	RU/mL	< 20
	IgM	6.32	RU/mL	< 20
Beta-2 glycoprotein		Negative		
Lupus anticoagulant		Negative		
Factor V Leiden		No mutation		
Mutation of prothrombin (20210 G-A)		No mutation		
Tests for HIV, VDRL and Lyme disease		Negative		
ANA, ANCA, dsDNA antibodies		Negative		

was mechanically ventilated (percutaneous tracheostomy was performed). Catecholamines and anti-oedematous treatment were used.

Seeking the cause of the stroke, vascular malformations were excluded via CT angiography. Transthoracic echocardiography was normal. Similarly, ultrasonographic examinations of the vessels of the lower limbs and abdominal cavity did not reveal any pathology, including thrombosis. In the diagnostic considerations, various causes of congenital and acquired thrombophilia were considered, including autoimmune diseases, and inflammatory factors. The activity of C-protein and factors VIII and IX were increased, possibly due to pregnancy. Several laboratory tests were performed, as set out in Table 1.

On the 56th day of her stay in hospital, the patient was transferred to the Neurological Rehabilitation Department (NRD). She was circulatory and respiratory-efficient, and showed symptoms of locked-in syndrome. She was conscious, non-verbal contact in the form of eyelid movements was possible, but with abolished eye movements, and features of

bulbar syndrome with paralysis of nerves IX, X, XII and V, VII on both sides and quadriplegia except for movement in the left foot. Paroxysmal crying and laughter were observed. The patient was fed through a gastric tube, and later a percutaneous endoscopic gastrostomy (PEG) was performed. In the NRD, doses of LMWH and ASA were continued until 32 weeks of pregnancy. Later, the dose of LMWH was doubled and ASA was discontinued. During hospitalisation, the patient was consulted many times by obstetricians and foetal vital signs were monitored.

In NRD, an extensive rehabilitation programme was introduced based on therapy according to the Bobath and PNF methods, starting with bedside rehabilitation. Breathing exercises, positioning, orofacial exercises, passive motor exercises, and classical massage were used. Thereafter, active exercises, motor coordination exercises, and general rehabilitation exercises were introduced. From a lying position, through gradual verticalisation, the patient was led to walking using a walker, initially with the assistance of a physiotherapist.

Speech therapy with PNF elements, some sensory integration, the Masako maneuver and Shaker exercises were introduced. After 94 days of hospitalisation, the tracheostomy tube was removed. The mobility of the limbs, swallowing and logical thinking improved. Semi-liquid oral nutrition was introduced, and then the physiological way of eating was gradually returned. After 111 days, the PEG was removed.

On the day of discharge from the NRD (after a 142-day stay in the NRD), the woman was conscious, being orally fed, in logical verbal contact, but emotionally unstable. Neurological examination revealed slight weakening of the muscle strength of the left limbs (grade 4 of the MRC scale) and cerebellar symptoms of ataxia, dysdiadochokinesia and disturbances of dynamic balance. The patient had gained the ability to feed herself and to move independently with the aid of a walker (NIHSS = 3 points, mRS = 1 point).

A caesarean section was performed in the 38th week of pregnancy. The newborn was in a good condition. The puerperium was uncomplicated.

15 months after the stroke, the presence of a patent foramen ovale (PFO) was diagnosed and a Septal Occluder was implanted (RoPE score = 9 points). The RoPE scale is used in patients who have had a cryptogenic stroke and who have been diagnosed with PFO. A result of 9 points (max = 10) confirms the need for an occluder (which was also performed on the patient after delivery). Head MRI revealed malacic lesions in the left cerebellar hemisphere, affecting also the left middle peduncle, in the central part of the pons and the medulla oblongata (Fig. 1D). Outpatient tests for thrombophilia were repeated and were negative.

At the time of writing, the patient is moving independently, without assistance. The latest neurological examination has shown minor deficits i.e. mild ataxia in the left upper limb, and tandem gait slightly disturbed (NIHSS = 1 point, mRS = 1 point). The woman has got married, and is taking care of her two children on a daily basis.

If an acute stroke is diagnosed in a pregnant woman, reperfusion treatment should be seriously considered. However, each such case should be treated individually, considering the risk of intrauterine bleeding [2, 3].

The presented case of a severe ischaemic stroke in a young pregnant woman with PFO in whom neither thrombolysis nor thrombectomy could be performed, shows that the neuroplasticity of a young person's brain and many months of arduous, comprehensive rehabilitation can give unexpectedly good results [4, 5]. Our presented case suggests that pregnancy may promote repair mechanisms after stroke.

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References

1. Halan T, Ortiz JF, Reddy D, et al. Locked-In Syndrome: A Systematic Review of Long-Term Management and Prognosis. Cureus. 2021; 13(7): e16727, doi: 10.7759/cureus.16727, indexed in Pubmed: 34471579.
2. Jermakow N, Maluchnik M, Sienkiewicz-Jarosz H, et al. Trends of stroke hospitalisation and fatality rates in young vs. elderly people in Poland during 2010-2019 decade. Neurol Neurochir Pol. 2022; 56(5): 417–427, doi: 10.5603/PJNNS.a2022.0055, indexed in Pubmed: 35900165.
3. Kremer C, Gdovinova Z, Bejot Y, et al. European Stroke Organisation guidelines on stroke in women: Management of menopause, pregnancy and postpartum. Eur Stroke J. 2022; 7(2): I–XIX, doi: 10.1177/23969873221078696, indexed in Pubmed: 35647308.
4. Karliński M, Kozera-Strzelińska D, Sienkiewicz-Jarosz H, et al. Reliability of prehospital diagnosis of acute cerebrovascular accident. Neurol Neurochir Pol. 2022; 56(1): 89–95, doi: 10.5603/PJNNS.a2022.0011, indexed in Pubmed: 35089594.
5. Wrona P, Wróbel D, Popadynets V, et al. Predictors of good functional outcome in ischaemic stroke patients without delayed neurological improvement after mechanical thrombectomy. Neurol Neurochir Pol. 2024; 58(2): 185–192, doi: 10.5603/pjnns.98219, indexed in Pubmed: 38324116.