The Egyptian Journal of Neurology, Psychiatry and Neurosurgery

CASE REPORT

Open Access

Generalized dystonia following resuscitation from a cardiac arrest: a case report and review of the literature

Ahmadou Bamba Mbodji[1*], Maouly Fall[2], Ndiaga Matar Gaye[1], Mamadou Ka[1], Jean Claude Dione[3], Moustapha Ndiaye[1] and Amadou Gallo Diop[1]

Abstract

Background A wide variety of movement disorders can be observed after cerebral hypoxia, including akineto-hypertonic syndrome and dystonia. Post-anoxic dystonia is a rare clinical syndrome that is not widely reported in the literatures. It is thought to be related to cerebral hypoxia leading to ischaemia of the basal ganglia.

Case description We report a case of an 11-year-old girl who represented with generalized dystonia following resuscitation from a cardiac arrest after open heart surgery. Brain MRI showed basal ganglia hypersignals in T2-FLAIR (fluid attenuated inversion recovery) weighted sequence and in the diffusion sequence without restriction of ACD in favour of subacute ischemic lesions. Treated with oral baclofen, the evolution was favourable with regression of the dystonia.

Conclusion It is often difficult to accurately predict the final neurological outcome of a patient who has survived cardiac arrest. Baclofen and anticholinergic can be used for the treatment for dystonia post-cerebral hypoxia.

Keywords Generalized dystonia, Cardiac arrest, Cerebral hypoxia

Introduction

Movement disorders following resuscitation from a cardiac arrest are varied. They are represented by parkinsonian syndrome, dystonia, chorea, tics, athetosis, tremor, and myoclonus [1, 2]. These disorders are rare and most often related to lesions of the basal ganglia, which are very sensitive to cerebral hypoxia [2, 3]. We describe a case of generalized dystonia secondary to cardiac arrest.

*Correspondence:
Ahmadou Bamba Mbodji
mbodjahmadoubamba@gmail.com
[1] Neurology Department, Fann University Hospital Center, Dakar, Senegal
[2] Neurology Department, Pikine Hospital, Dakar, Senegal
[3] Cardiovascular and thoracic surgery department, Fann, Dakar, Senegal

Case presentation

A 11-year-old child, had mitral and tricuspid valve disease for which an open heart surgery was performed on September 22nd, 2021. Immediately after the surgery, she had a haemorrhagic shock followed by a cardiorespiratory arrest requiring intensive resuscitation for 10 min which was successful. A week later, she developed intermittent cramps followed by generalized muscle contractions affecting the trunk and all 4 limbs (Additional file 1: video 1 and Additional file 2: video 2). The involvement was predominantly in the upper limbs and involved mainly twisting movements. The rest of the neurological examination was normal. Brain magnetic resonance imaging (MRI) showed basal ganglia hypersignals in attenuated fluid inversion recovery (T2-FLAIR) weighted sequence and in the diffusion sequence without restriction of apparent coefficient diffusion (ADC) in favour of subacute ischemic lesions (Figs. 1 and 2). Apart from a leucocytosis of 11,890/mm^3 and an anaemia of 10.2 g/

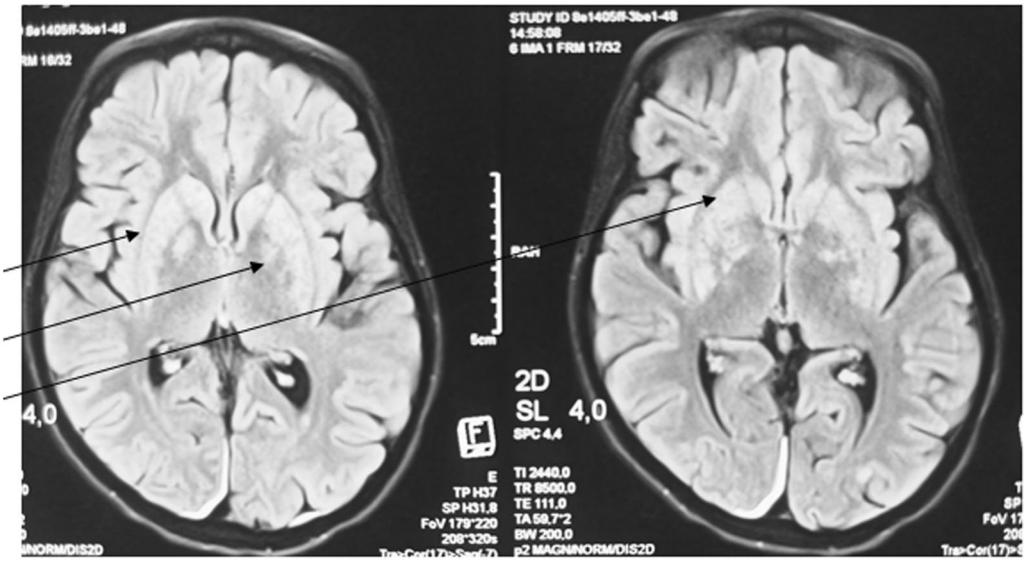

Fig. 1 Brain MRI in FLAIR sequence showing bilateral and symmetrical hypersignals of the basal ganglia (black arrow)

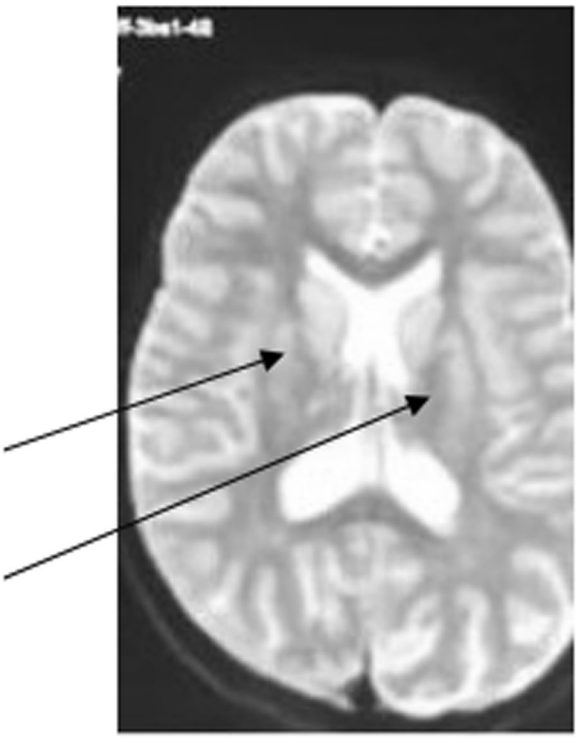

Fig. 2 Bilateral and symmetrical basal ganglia hypersignals on T2 sequences (black arrow)

dL, the rest of the blood exams were normal: sodium, potassium, calcium, chloride, carbon dioxide, blood urea nitrogen and creatinine. Blood toxicology screen tests were not performed. Oral medical treatment with baclofen (20 mg/day) was proposed. At the 2nd month of follow-up, we noted a regression of dystonia at the second month (Additional file 3: video 3).

Discussion

The clinical case we reported corresponds to a generalized dystonia secondary to a cardiac arrest in an 11-year-old girl. One of the particularities of this case was its rarity and its acute onset. Indeed, there are few cases described in the literatures [2, 4, 5]. A wide variety of movement disorders can be observed after cerebral hypoxia, including akineto-hypertonic syndrome and dystonia [3–5]. This latter may be focal, affecting a hand, foot, or cranial region, segmental or even generalized [2]. These disorders can occur acutely or with delay [7]. Indeed, in his review, Bhatt et al. found a delay in the onset of signs ranging from 1 week to 36 months after cardiac arrest [4]. There is an age-related difference in relation to the type of abnormal movements. The akineto-hypertonic syndrome is more frequent in the elderly, whereas dystonia is more frequent in young subjects such as our patient [2, 4–6]. However, the study of Scheibe et al. [8] have not confirmed this effect of age in the type of abnormal movements occurring after cardiac arrest. This could be related to the predominant inclusion of adults and the sample size in his study [8]. Pathophysiologically, it is generally accepted that lesions of the putamen are more responsible for dystonia, whereas lesions of the pallidum lead to an akineto-hypertonic syndrome [6, 7, 9]. However, cases of dystonia secondary to lesions of the medial pallidum have been reported in the literature [9, 10]. This could explain the clinical manifestations of our patient who presented with ischaemia of

the putamen and pallidum. The vulnerability of the basal ganglia to cerebral hypoxia/anoxia has been reported by several authors with different hypotheses, the main one being their terminal vascularization [4, 10]. The treatment of dystonia induced by cerebral hypoxia/anoxia is symptomatic [1]. Our patient received oral baclofen because anticholinergics were not available. Her evolution was favourable with a clear regression of dystonic movements. On the other hand, Wiltshire et al. and Ray et al. [11] did not find this same evolution and used other therapeutic means in association with baclofen [2].

Conclusion

It is often difficult to accurately predict the final neurological outcome of a patient who has survived cardiac arrest. Although rare, dystonias are among the complications secondary to cerebral hypoxia and which can alter the quality of life of patients. Their time to onset and evolution are variable. Their treatment is essentially symptomatic.

Abbreviations

FLAIR	Fluid attenuated inversion recovery
MRI	Magnetic resonance imaging
DWI	Diffusion-weighted imaging
ACD	Apparent coefficient diffusion

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s41983-023-00766-9.

Additional file 1. Video 1.

Additional file 2. Video 2.

Additional file 3. Video 3.

Acknowledgements
Not applicable.
Guarantor—Ahmadou Bamba Mbodji.

Author contributions
ABM, MF, NMG, MK, JCD, MN and AGD: these authors participated in the making and correction of this document. All authors agreed with the publication of the document.

Funding
The authors declare they have received no funding for the preparation of this document.

Availability of data and materials
Not applicable.

Declarations

Ethics approval and consent to participate
The ethical committee of the hospital gave the agreement to this study.

Consent for publication
Written informed consent was obtained from the patient's parent for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Competing interests
The authors declare that they have no competing interests.

Received: 4 November 2022 Accepted: 24 November 2023
Published online: 08 December 2023

References

1. Mehanna R, Jankovic J. Movement disorders in cerebrovascular disease. Lancet Neurol. 2013;12(6):597–608.
2. Wiltshire KM, Myers K, Wei X-C, Mohamed I. Asphyxiation causing distinctive basal ganglia injury and generalized dystonia. Can J Neurol Sci. 2011;38(3):514–5.
3. Hawker K, Lang AE. Hypoxic-ischemic damage of the basal ganglia. Case reports and a review of the literature. Mov Disord. 1990;5(3):219–24.
4. Bhatt MH, Obeso JA, Marsden CD. Time course of postanoxic akinetic rigid and dystonic syndromes. Neurology. 1993;43(2):314–7.
5. Venkatesan A, Frucht S. Movement disorders after resuscitation from cardiac arrest. Neurol Clin. 2006;24(1):123–32.
6. Bhatia KP, Marsden CD. The behavioural and motor consequences of focal lesions of the basal ganglia in man. Brain. 1994;117(Pt 4):859–76.
7. Burke RE, Fahn S, Gold AP. Delayed-onset dystonia in patients with static encephalopathy. J Neurol Neurosurg Psychiatry. 1980;43(9):789–97.
8. Scheibe F, Neumann WJ, Lange C, Scheel M, Furth C, Köhnlein M, et al. Movement disorders after hypoxic brain injury following cardiac arrest in adults. Eur J Neurol. 2020;27(10):1937–47.
9. Münchau A, Mathen D, Cox T, Quinn NP, Marsden CD, Bhatia KP. Unilateral lesions of the globus pallidus: report of four patients presenting with focal or segmental dystonia. J Neurol Neurosurg Psychiatry. 2000;69(4):494–8.
10. Kuoppamäki M, Bhatia KP, Quinn N. Progressive delayed-onset dystonia after cerebral anoxic insult in adults. Mov Disord. 2002;17(6):1345–9.
11. Ray S, Sidhu RJS, Pal PK. Status dystonicus as an acute sequelae following anoxic cerebral damage. Tremor Other Hyperkinet Mov (N Y). 2019;15(9):1–4. https://doi.org/10.5334/tohm.564.

Publisher's Note