

17 - 2020 - Successful Relief of Abdominal Dystonia After Sequential GPi Pallidotomy with 2-Year Follow-Up

by Achmad Fahmi

Submission date: 13-Sep-2022 03:47PM (UTC+0800)

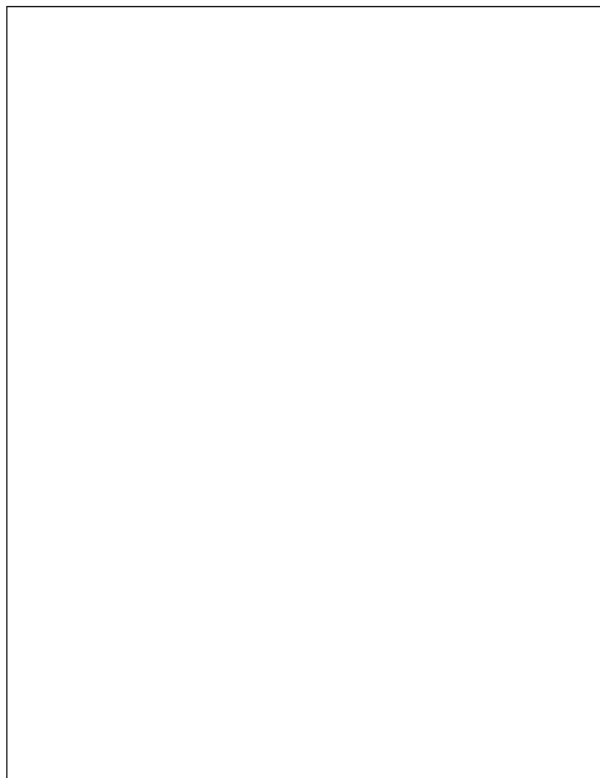
Submission ID: 1898698510

File name: tonia_After_Sequential_GPi_Pallidotomy_with_2-Year_Follow-Up.pdf (1.34M)

Word count: 1987

Character count: 11514

Provided for non-commercial research and education use.
Not for reproduction, distribution or commercial use.



This article appeared in a journal published by Elsevier. The attached copy is furnished to the author for internal non-commercial research and education use, including for instruction at the author's institution and sharing with colleagues.

Other uses, including reproduction and distribution, or selling or licensing copies, or posting to personal, institutional or third party websites are prohibited.

In most cases authors are permitted to post their version of the article (e.g. in Word or Tex form) to their personal website or institutional repository. Authors requiring further information regarding Elsevier's archiving and manuscript policies are encouraged to visit:

<http://www.elsevier.com/authorsrights>



6 Successful Relief of Abdominal Dystonia After Sequential GPi Pallidotomy with 2-Year Follow-Up

Achmad Fahmi^{1,3}, Asadullah^{2,3}, Yunus Kuntawi Aji^{2,3}, Dirga Rachmad Aprianto^{2,3}, Heri Subianto^{2,3}, Agus Turchan^{2,3}

Key words

- Abdominal dystonia
- GPi pallidotomy

Abbreviations and Acronyms

GPi: Globus pallidus internus

From the ¹Post Graduate Doctoral Program, Faculty of Medicine and ²Department of Neurosurgery, Faculty of Medicine, Universitas Airlangga, Surabaya; and ³Dc. Soetomo General Academic Hospital, Surabaya, Indonesia

To whom correspondence should be addressed:

Achmad Fahmi, M.D., Ph.D.

[E-mail: achmad.fahmi-13@fk.unair.ac.id]

Supplementary digital content available online.

Citation: *World Neurosurg.* (2020) 144:68-70.

<https://doi.org/10.1016/j.wneu.2020.08.152>

Journal homepage: www.journals.elsevier.com/world-neurosurgery

Available online: www.sciencedirect.com

1878-8750/\$ - see front matter © 2020 Elsevier Inc. All rights reserved.

INTRODUCTION

Abdominal dystonia, a form of focal dyskinesia affecting the abdominal wall,¹ was first described by Antonie Van Leeuwenhoek in 1723 when he himself experienced symptoms.² Since then, various terms have been proposed for this disorder, including Leeuwenhoek's disease, diaphragmatic myoclonus, diaphragmatic tremor, diaphragmatic flutter, moving umbilicus dyskinesia, abdominal dystonia, and respiratory myoclonus.³⁻⁵

Abdominal dystonia is extremely rare, consisting of involuntary and repetitive rhythmic movements of the abdominal wall. These movements cannot be suppressed voluntarily, but may be influenced by respiratory maneuvers. Investigations, such as spinal cord and abdominal imaging, usually fail to reveal any local abnormalities to explain this movement disorder.^{6,7} Various cases have been reported in the literature with a long list of underlying causes. However,

BACKGROUND: Abdominal dystonia is very rare. To our knowledge, no clinical study has reported its specific treatment. Stereotactic therapy has been used to treat several movement disorders, including focal and general dystonia. We investigated the use of internal globus pallidum (GPi) pallidotomy for abdominal dystonia after failed oral medication.

CASE DESCRIPTION: A 48-year-old man presented with abdominal dystonia and complaints of involuntary undulating and contraction movements of his left abdominal wall for 5 years. Treatment with oral medication for 4 years was ineffective. Lesioning of the right GPi successfully relieved his symptoms. The symptoms recurred at 3 months and right GPi pallidotomy was repeated with complete resolution of symptoms after the second procedure. There was no recurrence or focal deficit at the 2-year follow-up.

CONCLUSIONS: GPi pallidotomy is feasible and effective for the treatment of abdominal dystonia that is resistant to standard medical therapy.

to our knowledge, the exact underlying pathophysiology has not been elucidated. In such cases, symptomatic management is the only available option.⁷ The prognosis is unfavorable because no effective treatment exists.⁸ We reported our experience with a case of abdominal dystonia treated with internal globus pallidus (GPi) pallidotomy and stereotactic lesioning. A 2-year follow-up is reported.

CASE PRESENTATION

A 48-year-old man presented to our hospital with a 5-year history of involuntary jerking movements of the left anterior abdominal wall. The symptoms gradually worsened with time. The involuntary movements occurred suddenly without any associated aura or obvious trigger. The patient felt the abdominal muscle contractions especially on the left side (see [Video 1](#)). The contraction and jerking movements did not improve with resting, sitting, or lying positions. The tightness and pain felt on the abdominal wall caused work and sleep disturbances. The patient had no

history of medication use or previous trauma to the abdomen and head. There was no family history of this condition. Neurologic examination and magnetic resonance imaging showed no abnormalities that could explain the symptoms.

The patient received oral medication (including clonazepam, trihexyphenidyl, diazepam, and baclofen) for 4 years without relief. Diagnosis was abdominal dystonia resistant to oral medication. After a brief discussion between the team and patient about any other modality including botulinum toxin injection and surgery, we performed stereotactic right GPi pallidotomy ([Figure 1](#)). The symptoms subsequently resolved completely, and the patient was discharged without any medication. The symptoms recurred after 3 months, and GPi pallidotomy was repeated 3 months later (6 months after the initial pallidotomy) according to the same technique, but in different target locations.

Lesioning was performed using an Inomed ZD (Inomed, Emmendingen, Germany) stereotactic frame. The right GPi target was set at 20.5 mm lateral, 4.0



Video available at www.sciencedirect.com

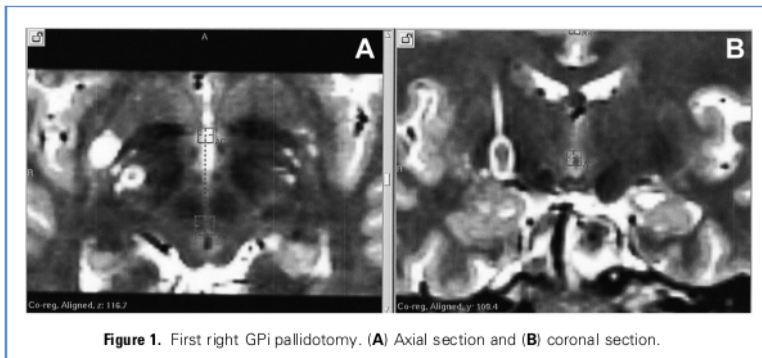


Figure 1. First right GPi pallidotomy. (A) Axial section and (B) coronal section.

10 mm inferior, and 2.5 mm anterior from the midpoint of the anterior-posterior commissures, 3 mm 45° anteromedial from the center of the first pallidotomy lesion. We used a 1.1-mm-diameter electrode with a 4-mm active tip. Cosman radiofrequency was used to create a lesion via thermocoagulation at 70°C for 30 seconds (Figure 2). After the second pallidotomy, the symptom resolved completely. The Burke-Fahn-Marsden dystonia rating scale scores were

significantly improved from 12 to 0 compared with that before the second GPi pallidotomy, the patient remained in good condition with no symptoms or neurologic deficit at 2 years of follow-up (see Video 2).

DISCUSSION

Our patient, who had a 5-year history of abdominal dystonia, had intractable symptoms even after multimodal medical

therapy. Taira et al⁹ noted that most cases of dystonia are medically refractory and that surgical treatment results in marked improvement. From physical examination, laboratory findings, and head magnetic resonance imaging, we believed that the etiology in our patient was idiopathic. Krack and Vercueil¹⁰ reported great benefit from surgical treatment, especially in cases of primary (idiopathic or genetic) dystonia and less so with secondary dystonia.

Recently, various neurosurgical interventions have been effective for various types of dystonia.⁹ Based on some recent series of patients, lesioning in the GPi has been currently safe and may be the best treatment for dystonia.^{10,11} This statement was further supported by Zhuang et al,¹² who reported that neurons in 87% of 367 patients with dystonia exhibited altered neural activity, including grouped discharge in the GPi and subthalamic nucleus, long-lasting neuronal activity, and rapid neuronal discharge in the ventralis oralis posterior or ventralis intermediate nucleus of the thalamus. Neurons in the ventralis oralis posterior, GPi, and subthalamic nucleus were firing at the same frequency as on electromyography during dystonia.¹² Yoshor et al¹³ recommended GPi pallidotomy as optimal therapy for most patients with primary dystonia.

Presently, although lesioning has been replaced primary by deep brain stimulation due to its greater safety and the ability to perform adjustable lesions, as Taira et al¹⁴ described and has been shown to improve dystonia,¹⁵ we believed that lesioning remains a good treatment modality for patients who are unwilling to undergo device implantation and/or who cannot undergo deep brain stimulation for economic or geographic reasons. After a brief discussion, our patient underwent lesioning in the right GPi with excellent improvement immediately postoperatively.

The symptoms recurred 3 months postoperatively. After a brief discussion, supported by the study of Krystkowiak et al¹⁶ in 5 patients with focal dystonia, we suggested that the recurrent symptoms may be caused by altered neuronal activity in the right GPi interrupting the cortico-striato-pallido-thalamo-cortical loop induced by the lesion we created at the initial

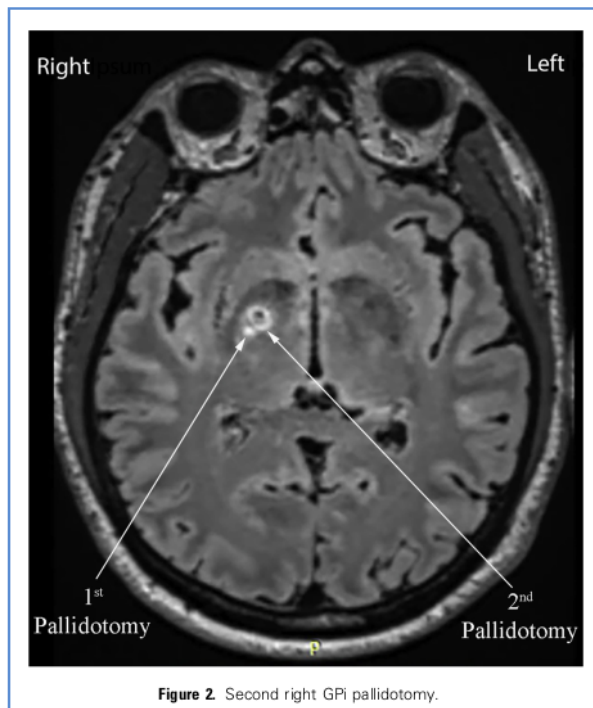


Figure 2. Second right GPi pallidotomy.

pallidotomy. From another study, we concluded that this type of dystonia responds poorly to most medical therapies and surgical therapy yielded the best result.¹⁷ We planned a sequential pallidotomy in the right GPI and symptoms resolved completely after the second procedure. At 2-year follow-up, there was no recurrence or neurologic deficit.

In conclusion, we reported a rare case of abdominal dystonia treated successfully with GPI pallidotomy. This case indicates that GPI pallidotomy can be a safe and effective treatment of choice for patients with abdominal dystonia resistant to medication or those who could not tolerate the medication side effects.

ACKNOWLEDGMENTS

We acknowledge Dr. Soetomo General Academic Hospital and National Hospital, Surabaya, Indonesia, where the surgery was performed. We would also like to thank Enago (www.enago.com) for the English language review.

REFERENCES

- Bhidayasiri R, Tarsy D. Belly dancer's dyskinesia. In: Bhidayasiri R, Tarsy D, eds. *Movement Disorders: A Video Atlas*. Totowa, New Jersey: Humana Press; 2012:176-177.
- Lamer AJ. Antony van Leeuwenhoek and the description of diaphragmatic flutter (respiratory myoclonus). *Mov Disord*. 2005;20:917-918.
- Patterson V. Belly dancer's syndrome: causes, clinical presentations, and treatment options. Published online; 2011. <https://www.logan.edu/mm/files/LRC/Senior-Research/2011-Dec-31.pdf>. Accessed August 6, 2020.
- Maramattom BV, Sreekumar P, Kumar RA. Diaphragmatic flutter masquerading as palpitations. *Ann Indian Acad Neurol*. 2016;19:402-403.
- Ramirez JD, Gonzales M, Hoyos JA, Grisales L. Diaphragmatic flutter: a case report and literature review. *Neurologia*. 2015;30:249-251.
- Meyer JA, Desai KV, Geyer HL. Recurrent belly dancer dyskinesia in pregnancy. *Neurology*. 2017; 88:2066.
- Gupta A, Kushwaha S. Belly dancer's dyskinesia: a glimpse of a rare phenomenon. *Cureus*. 2017;9: e1457.
- Linazasoro G, Van Blercom N, Lasa A, Fernández JM, Aranzábal I. Etiological and therapeutic observations in a case of belly dancer's dyskinesia. *Mov Disord*. 2005;20:251-253.
- Taira T, Ochiai T, Goto S, Hori T. Multimodal neurosurgical strategies for the management of dystonias. *Acta Neurochir Suppl*. 2006;99: 29-31.
- Krack P, Vercueil L. Review of the functional surgical treatment of dystonia. *Eur J Neurol*. 2001;8: 389-399.
- Shetty AS, Bhatia KP, Lang AE. Dystonia and Parkinson's disease: What is the relationship? *Neurobiol Dis*. 2019;132:104462.
- Zhuang P, Li Y, Hallett M. Neuronal activity in the basal ganglia and thalamus in patients with dystonia. *Clin Neurophysiol*. 2004;115: 2542-2557.
- Yoshor D, Hamilton WJ, Desaloms JM, Ondo W, Jankovic J, Grossman RG. Stereotactic neurosurgery for dystonia. *Mov Disord Surg*. 2004;15:279-295.
- Horisawa S, Nanke M, Kawamata T, Taira T. Pallidothalamic tractotomy for Parkinson disease with 1-year follow-up: a case report. *World Neurosurg*. 2019;121:193-195.
- Ehrlich DJ, Frucht SJ. The phenomenology and treatment of idiopathic adult-onset truncal dystonia: a retrospective review. *J Clin Mov Disord*. 2016; 3:15.
- Krystkowiak P, Martinat P, Defebvre L, Pruvo JP, Leys D, Destée A. Dystonia after striatopallidal and thalamic stroke: clinicoradiological correlations and pathophysiological mechanisms. *J Neurol Neurosurg Psychiatry*. 1998; 65:703-708.
- Chuang C, Fahn S, Frucht SJ. The natural history and treatment of acquired hemidystonia: report of 33 cases and review of the literature. *J Neurol Neurosurg Psychiatry*. 2002;72:59-67.

Conflict of interest statement: The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Received 20 June 2020; accepted 22 August 2020

Citation: *World Neurosurg*. (2020) 144:68-70. <https://doi.org/10.1016/j.wneu.2020.08.152>

Journal homepage: www.journals.elsevier.com/world-neurosurgery

Available online: www.sciencedirect.com

1878-8750/\$ - see front matter © 2020 Elsevier Inc. All rights reserved.

17 - 2020 - Successful Relief of Abdominal Dystonia After Sequential GPi Pallidotomy with 2-Year Follow-Up

ORIGINALITY REPORT

18%

SIMILARITY INDEX

12%

INTERNET SOURCES

14%

PUBLICATIONS

1%

STUDENT PAPERS

PRIMARY SOURCES

- | | | |
|---|--|----|
| 1 | coek.info
Internet Source | 2% |
| 2 | Zhuang, P.. "Neuronal activity in the basal ganglia and thalamus in patients with dystonia", <i>Clinical Neurophysiology</i> , 200411
Publication | 2% |
| 3 | Achmad Fahmi, Heri Subianto, Agus Turchan. "Commentary: Pre- and Postoperative Gait Analysis and Video for Selective Dorsal Rhizotomy in Spastic Diplegia: 2-Dimensional Operative Video", <i>Operative Neurosurgery</i> , 2019
Publication | 2% |
| 4 | Shiro Horisawa, Magi Nanke, Takakazu Kawamata, Takaomi Taira. "Pallidothalamic Tractotomy for Parkinson Disease with 1-Year Follow-Up: A Case Report", <i>World Neurosurgery</i> , 2019
Publication | 1% |
| 5 | www.cern.ac.cn | |

Internet Source

1 %

6

www.annalsofian.org

Internet Source

1 %

7

journals.plos.org

Internet Source

1 %

8

"Abstracts", Movement Disorders, 2019

Publication

1 %

9

kenanarnautovicmd.com

Internet Source

1 %

10

academic.oup.com

Internet Source

1 %

11

goforexyourself.com

Internet Source

1 %

12

lbeifits.files.wordpress.com

Internet Source

1 %

13

Dunbar Alcindor, Michael Y. Oh, Susan Baser, Cindy Angle, Boyle C. Cheng, Donald Whiting. "Stimulation of the globus pallidus internus in a patient with DYT1-positive primary generalized dystonia: a 10-year follow-up", *Neurosurgical Focus*, 2010

Publication

1 %

14

P Krystkowiak, P Martinat, L Defebvre, J P Pruvo, D Leys, A Destee. "Dystonia after

1 %

striatopallidal and thalamic stroke: clinikoradiological correlations and pathophysiological mechanisms", Journal of Neurology, Neurosurgery & Psychiatry, 1998

Publication

15

movementdisorders.onlinelibrary.wiley.com

Internet Source

1 %

16

"Contents", World Neurosurgery, 2020

Publication

1 %

17

"Deep Brain Stimulation in Neurological and Psychiatric Disorders", Springer Science and Business Media LLC, 2008

Publication

1 %

18

doi.org

Internet Source

1 %

19

Dina Angelika, Risa Etika, Muhammad Pradhika Mapindra, Martono Tri Utomo, Paulus Rahardjo, I Dewa Gede Ugrasena. "Associated neonatal and maternal factors of osteopenia of prematurity in low resource setting: A cross-sectional study", Annals of Medicine and Surgery, 2021

Publication

<1 %

Exclude quotes On

Exclude matches Off

Exclude bibliography On

17 - 2020 - Successful Relief of Abdominal Dystonia After Sequential GPi Pallidotomy with 2-Year Follow-Up

GRADEMARK REPORT

FINAL GRADE

/100

GENERAL COMMENTS

Instructor

PAGE 1

PAGE 2

PAGE 3

PAGE 4
