International Journal of Surgery Case Reports 87 (2021) 106422

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Early internal capsule infarction following globus pallidus internus lesioning for cervical dystonia

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ARTICLE INFO

Keywords:
GPi Pallidotomy
Dystonia
Infarction
Complication
Ablation

ABSTRACT

Introduction and importance: Ischemia associated with lesioning for the treatment of cervical dystonia is a very rare phenomenon. We reported a case of early internal capsule infarction following GPi lesioning for cervical dystonia.

Case presentation: A 56-year-old man with one year history of progressive onset of cervical dystonia which was unresponsive to medical therapy. He was planned for bilateral globus pallidus internus pallidotomy. After completion of the procedure on one side and the successful completion of stimulation and test lesion on another side, the patient suddenly developed dysarthria and one sided weakness. Post-operative magnetic resonance imaging revealed acute infarction in the posterior end of the internal capsule. The patient was managed conservatively and underwent physiotherapy and rehabilitation care. He recovered gradually with modified ranking scale 3 on discharge.

Clinical discussion: Globus pallidus internus lesioning or ablation is one of the surgical treatment for dystonia and movement disorder. Though considered as a safe technique, various risk are associated with the procedure. Ischemia is one of the associated risk but is a very rare phenomenon. Though the patient did not have any signs of pre-operative ischemia, the occlusion of one of the perforators to internal capsule during ablation may be the cause of intra-operative ischemia.

Conclusion: Ischemia associated with lesioning in the treatment of cervical dystonia is a very rare phenomenon. Though, ischemia related with radiofrequency ablation is very rare and sporadic, one has to be very cautious during GPi pallidotomy to prevent occlusion of perforators to internal capsule.

1. Introduction

Cervical dystonia is related to involuntary neck movements like horizontal rotation, ante- and retroflection, and lateral bending that needs a complex combination of bilateral neck muscles [1]. Deep brain stimulation (DBS) is a commonly performed procedure for the treatment of cervical dystonia nowadays. However, lesioning of Globus pallidus internus (GPi) can be an option for the treatment of cervical dystonia [2]. Many complications have been reported regarding the treatment of movement disorders such as hemorrhage, infection, edema etc. [3,4] However, ischemia associated with DBS or lesioning is a very rare phenomenon. So, we reported a case of early internal capsule infarction following GPi lesioning for cervical dystonia. The case has been reported in accordance to the SCARE guideline [5].

2. Case report

A 56 year old man with 4 years history of progressive onset of cervical dystonia which was refractory to medical therapy (baclofen, clonazepam). He is a diagnosed case of hypertension for which he was under Amlodipine. He had no history of head trauma or other possible contributing factors. He had no significant family history. The Burke-Fahn-Marsden cervical dystonia rating on examination was found to be 36 [6].

On examination, he had spontaneous turning of head to right side. Tightness was noted on his right sternoclenoidmastoid (SCM) and right trapezium muscle, however, no muscle atrophy was noticed. For this he was planned for GPi pallidotomy and magnetic resonance imaging (MRI) of the brain was done, which revealed that the brain was grossly

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https://doi.org/10.1016/j.ijscr.2021.106422
Received 23 August 2021; Received in revised form 13 September 2021; Accepted 14 September 2021
Available online 16 September 2021

intact.

As a part of the protocol, brain MRI (3 T, Siemens) with no spacing and 3-Dimensional Volume reconstruction image was performed and it was obtained in digital imaging communication in medicine (DICOM) standard. The image was loaded in Inomed Planning Software (IPS), where the targeting and trajectory for stereotactic GPi pallidotomy was done (Fig. 1). The anterior commissure (AC) and posterior commissure (PC) were visualized and AC- PC distance was calculated to be 24.2 mm. The primary target on the right side was set as 19.5 mm lateral, 3 mm below (caudal) and 2 mm anterior to the mid-commissural point and the target on the left side was set as 19.5 mm lateral, 3 mm below and 2 mm anterior to the mid-commissural point. Similarly, secondary target was set for both right and left side as 21 mm lateral, 1 mm posterior and 3 mm below the mid-commissural line. The target was reconfirmed through the inbuilt Schaltenbrand atlas (Fig. 2). Under local anesthesia, stereotactic frame (MRI compatible Zamorano-Dujovny Fisher) frame was applied and computed tomography (CT) scan of head was done (64 slice Siemens with 2 mm slice thickness) with no tilt. These images were retrieved in a DICOM format. Then the MRI and CT images were fused in the workstation and the working coordinates for the surgical procedure was extracted. The patient was then taken to the operating room and the frame was fixed in the Mayfield. The coordinates were set on phantom to reconfirm the accuracy of target. Once confirmed, a burr hole just anterior to coronal suture was made.

First, the procedure was done on the left side. Radio frequency (RF) electrode was inserted and the correctness of the placement was checked with c-arm (fluoroscopy) image. Stimulation was done at 50 Hz/0.1 msec/2.5 mA (frequency/pulse width/amplitude). No deficit was noted, so the test lesion was made at 50 °C for 30s at 3 mm below target, which partially eased the patient's cervical tightness without any other side-effects. Hence, a permanent lesion was made with 75 °C for 60s at +3, +2, +1, 0, −1, −2 positions in relation to the target. No complication or deficit was noted while creating the permanent lesion. Then, secondary lesion was planned. Stimulation and test lesion with above mentioned parameters did not show any complications at 3 mm deeper to the target. But unfortunately while a creating lesion at +2, patient suddenly complained of dysarthria and right sided weakness. The procedure was promptly stopped and wound was closed in layers. Immediate CT was

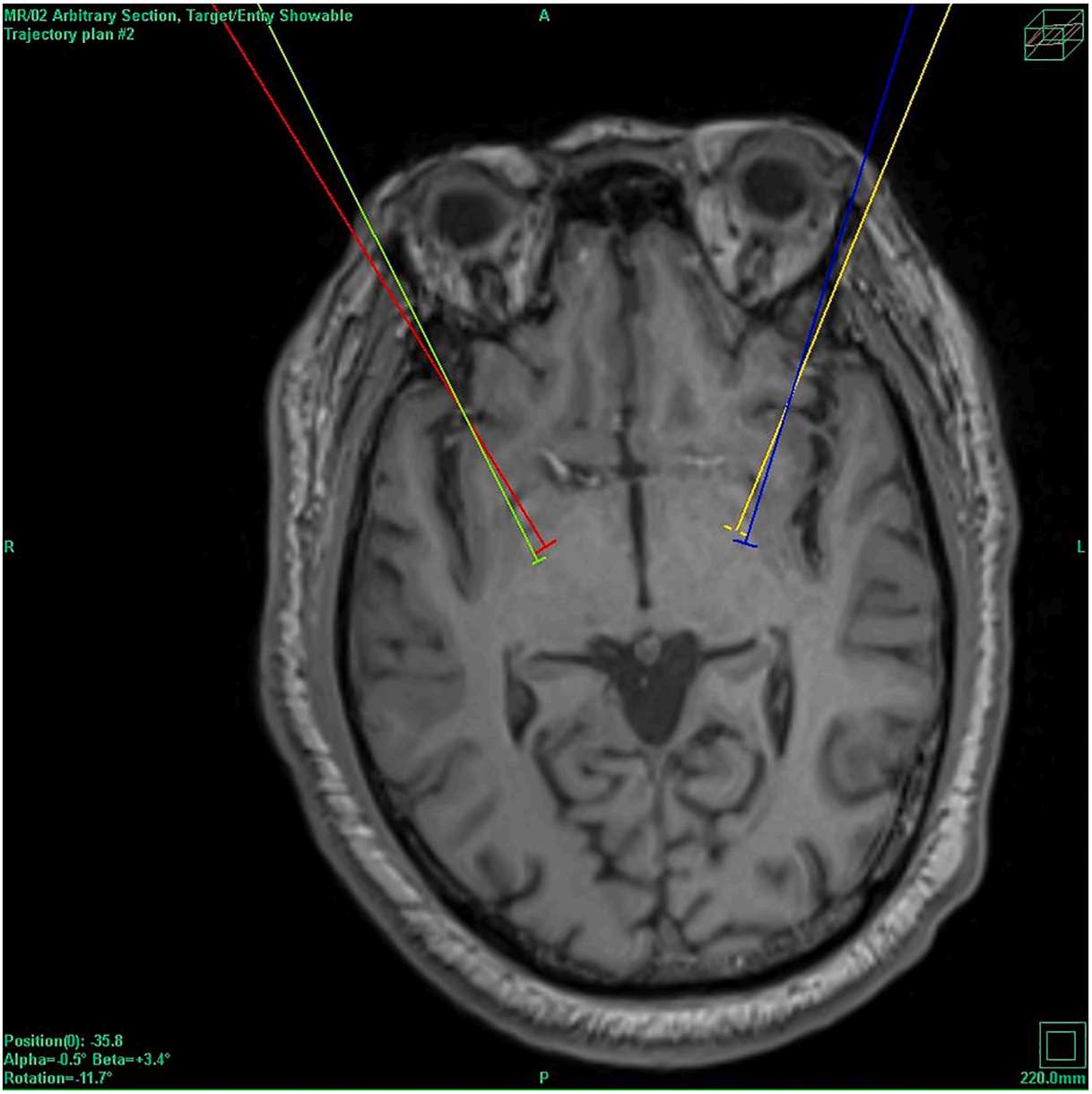

Fig. 1. Target set for GPI Pallidotomy in IPS software. Axial MRI Image.

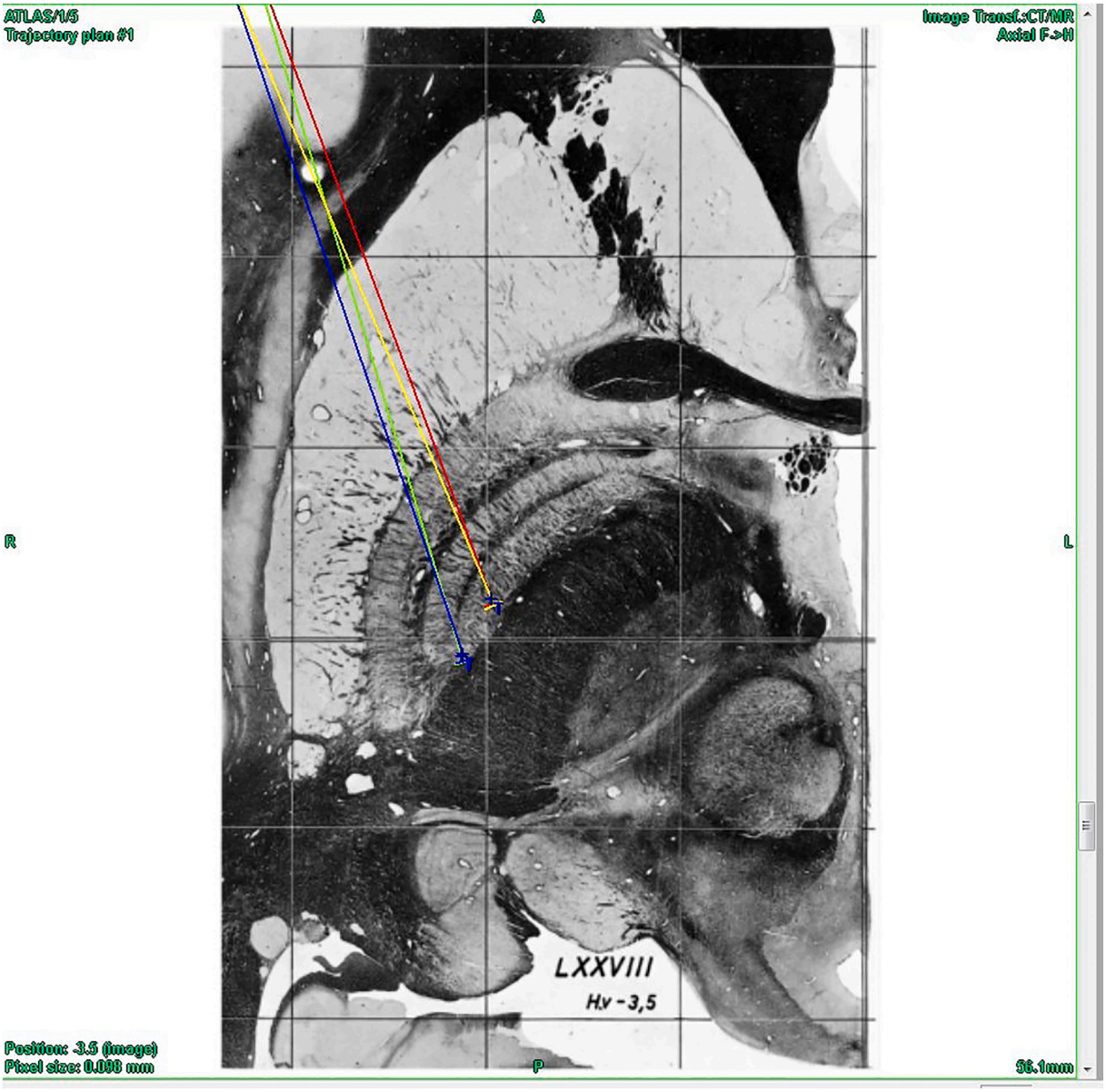

Fig. 2. Target confirmed in Schaltenbrand Atlas (axial view).

performed which revealed no acute blood. On the next day, MRI was performed which revealed acute infarction in the posterior end of the internal capsule (Fig. 3). The patient was managed conservatively, where he underwent physiotherapy and rehabilitation care. He had recovered gradually with modified ranking scale (mRS) 3 on discharge after 10 days of surgery.

On follow up after three months of surgery, the patient had significant improvement in cervical dystonia with Burke-Fahn-Marsden score of 10. However, the effect of ischemia was still persistent but slightly improved than before with motor power of lower limb 4/5 and upper limb 2/5, with weak hand grip.

3. Discussion

Various treatment modalities have been found for the treatment of movement disorders from invasive to less invasive such as chemical ablation using alcohol, radiofrequency lesioning, gamma knife radiosurgery, deep brain stimulation and magnetic resonance guided focused ultrasound surgery (MRgFUS) [7–9]. The complications of hemorrhage, infection, edema have been reported in various papers [3,4]. However, ischemia after the procedures for the treatment of movement disorder is very rare.

Review of literature revealed few papers associated with subcortical ischemia following DBS and lesioning [10,11]. Most of subcortical ischemia are seen in patients who undergo DBS, mostly during microelectrode recording (MER). Kang et al. reported an ischemic stroke of the posterior limb of the internal capsule during GPi DBS procedure in a 34- year-old patient with cervical dystonia [12]. Downes et al. reported the incidence of ischemic stroke during GPi DBS was 2.14% per lead [13]. They reported ischemic stroke in 5 cases. MER was used in all the cases. Among them only 3 patients experienced symptom onset during MER while in the other 2 cases, symptom onset was noted before the initiation of and during macro-stimulation. Novak reported 2 cases of the unusual adverse event of ischemia associated with DBS placement for advanced Parkinson's disease [14]. In the first case, the clinical effects of ischemia were observed intraoperatively which was supported by the silence (no signal) during microelectrode recording from the ischemic region. While, in the second case, the timing of the ischemic event could not be determined accurately but also was associated with a difficult mapping.

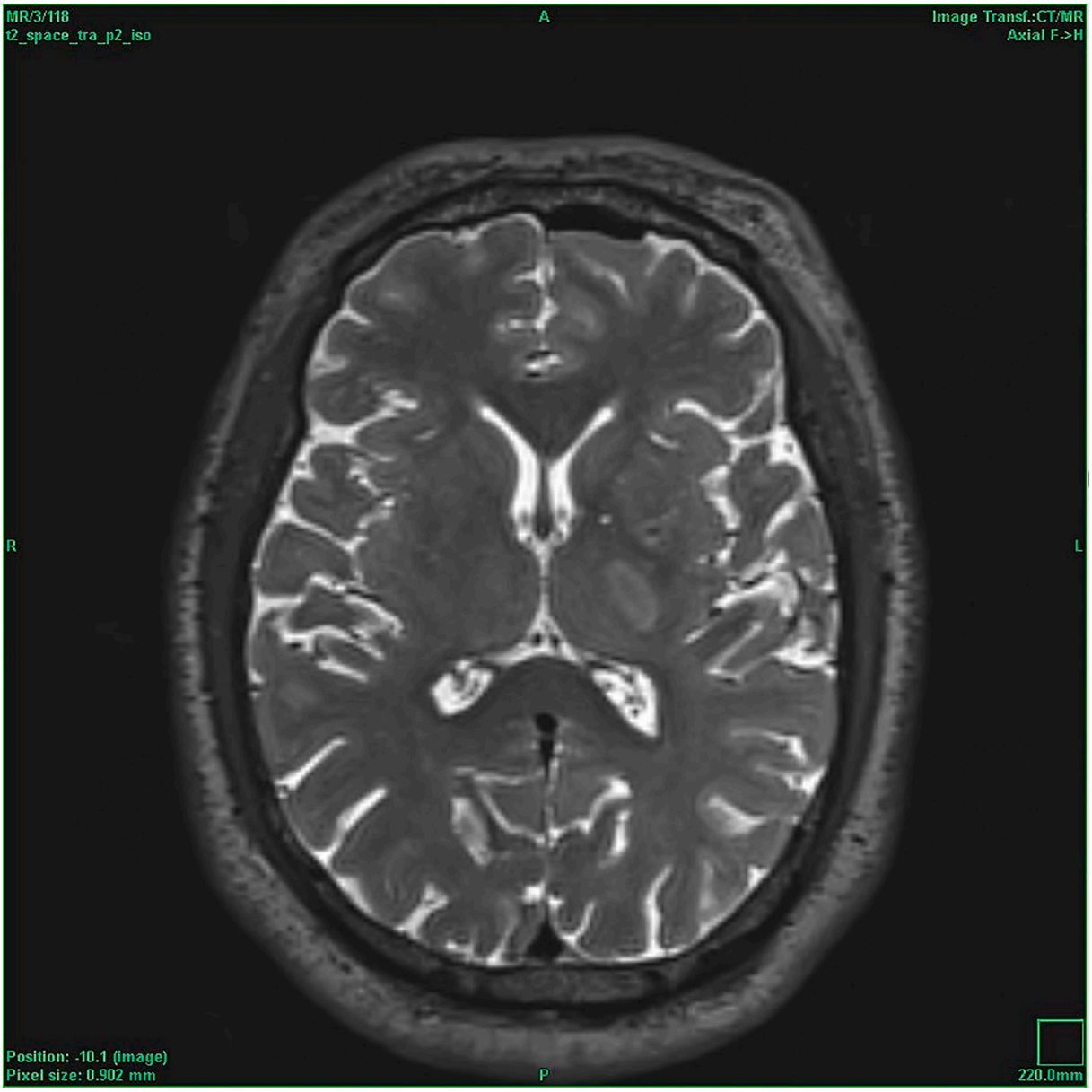

Fig. 3. Post-operative MRI showing a lesion in the left GPi and a small infarction in internal capsule.

There have been few papers of subcortical ischemia after GPi Pallidotomy. Christoforidis reported a case of a 54-year-old man with Parkinson's disease involving predominantly the right side, progressively deteriorating under medical management [10]. The patient developed right facial weakness and pronator drift during the procedure of stereotactically guided radiofrequency thermal ablation of the posteroventral globus pallidus internus. Lim reported 3 cases of delayed infarction in a series of patients with idiopathic Parkinson's disease (IPD) who underwent stereotactic radiofrequency (RF) pallidotomies [11]. All three patients had significant previous vascular disease and were in a high-risk group for cerebral infarction.

Lim et al. showed that there was a significantly higher incidence of infarction in patients with preexisting vascular disease compared to that in patients who only had risk factors for vascular disease [11]. Kang et al. said that one possible mechanism of ischemic stroke during movement disorder surgery is vasospasm induced by electrical stimulation [12]. Novak et al. believe that the area of edema may compress the small penetrating arteries, thus resulting in ischemia. While Christofordis et al. suggested that thrombosis or spasm of a lenticulostriate artery is the cause of ischemic infarction in the internal capsule [10]. Downes et al. said that it could be related to compression, or vasospasm of lenticulostriate arteries and/or anterior choroidal branches near the GPi target [13]. In our case, we believe that it could have occurred after the occlusion of perforator supplying the internal capsule while performing electrical stimulation or test lesion. The patient in our case did not have preexisting infarction but had known risk factor of hypertension.

In general, it is difficult to measure cerebral blood flow (CBF) during operation. However, near infrared spectroscopy (NIRS) or Laser speckle contrast imaging (LSCI) can be alternatives for measuring local CBF during operation to prevent ischemia. Unfortunately we did not use such modalities in our set up. In future the analysis of ischemia interacted with NIRS or LSCI would be productive complementarily.

4. Conclusion

Ischemia associated with lesioning in the treatment of movement disorder is a very rare phenomenon. Rare occurrence of such events

during lesioning makes it quite difficult to comprehend, however, such adverse effects are inevitable and one has to be very cautious about ischemia while performing lesioning for the movement disorder.

Funding

The case study was not funded by any institutes.

Ethical approval

The case study was cleared by the institutional review committee of Annapurna Neurological Institute and Allied Sciences.

Consent

A written and signed consent for publication has been received from the patient and his family.

Registration of research studies

Not applicable.

Guarantor

Resha Shrestha
Basant Pant

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Pritam Gurung: Study design, writing original draft, conceptualization and review.
Resha Shrestha: Study design and analysis.
Sambardhan Dabadi: review and editing.
Raju Raj Dhungel: review and editing.
Bishal Shrestha: Data analysis and writing.
Basant Pant: Supervision.

Declaration of competing interest

The authors have no conflicts to disclose.

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