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Case report: Dystonia and athetosis in ICH of 80 years old female patient

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ABSTRACT

Dystonia is defined as involuntary and persistent contraction of agonist and antagonist muscles resulting in abnormal posture, twisting and repetitive movements, or trembling. These can be initiated or worsened by attempts to make movements. Dystonia is a dynamic disorder whose severity changes based on activity and body posture. Case An 80 years-old woman was brought by her family to the emergency room at RAA Soewondo Regional Hospital, Pati with complaints of movements, such as twisting her arms and legs from the right arm and leg continuously without stopping which appeared since 2 (two) days before entering the hospital. Physical examination revealed continuous voluntary and twisting movements of the right arm and leg. There was a neurological deficit in nerve VII, namely; eyebrows, nasolabial sulcus, and corners of the mouth that deviate to the left. Moreover, there are also neurological deficits in nerve XII, namely; tongue deviation to the left side and tongue deviation during contraction to the right side. Therefore, patients receive inpatient treatment at the hospital. Conclusion: is dystonia is a clinical diagnosis made based on history, observation, and direct examination. Treatment provided depends on the patient's age, diagnosis, and distribution of dystonia, and is individualized, consisting of rehabilitation, botulinum toxin injections, oral pharmacotherapy, and DBS.

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INTRODUCTION

Dystonia, the third most common movement disorder after essential tremor and Parkinson's disease (PD), is characterized by agonist and antagonist muscle contractions resulting in abnormal posture and uncontrolled movements (Radhakrishnan & Goyal, 2018; Sukmawati, 2018; Wu & Hallett, 2013). Contractions can be continuous or intermittent (Hayes, 2019; Opara et al., 2017). Intermittent contractions can occur spontaneously or be task-dependent. Dystonia can affect any

part of the body, leading to varied manifestations (Cramb et al., 2023; Elbaz et al., 2016; Sveinbjornsdottir, 2016).

Most forms of dystonia are idiopathic. However, some genetic dystonias have been identified, and the acquired forms are usually associated with lesions in the basal ganglia or more global brain injury (Gudden et al., 2021; Kumar et al., 2014; Maretalinia et al., 2023; Matar et al., 2020). Pathophysiology Dystonia is currently thought to represent a brain tissue disorder involving multiple brain regions, including the basal ganglia, cerebellum, thalamus, and sensorimotor cortex, resulting in abnormal motor neuron programming. Treatment involves regular botulinum toxin injections, and oral medications. It is not reliable. Deep brain stimulation (DBS) can be very effective in certain cases (Connors et al., 2018; Faustina, 2008; Prasad et al., 2023; Scott et al., 2022; Sukmawati et al., 2023).

Since the heterogeneous nature of dystonia, and epidemiological data are limited, a meta-analysis demonstrated the overall prevalence of dystonia isolated focal is 16,4 per 100,000 people. Dystonia focal is more common than cases with a wider distribution (segmental, multifocal, and generalized). Although rarely happened in childhood, dystonia becomes more common with age. Prevalence rates of dystonia early onset is defined as onset before age 20 years, is 0,2 to 5 per 100.000. Compared with 3 to 732 per 100.000 for dystonia late onset is defined as onset at age 20 years or later. In a door-to-door study in India, the prevalence of dystonia in the age group less than 30 years was 7,7 per 100.000 population, while the prevalence of dystonia in the age group between 50 and 70 years was 177,9 per 100.000 population.

Among the adult-onset forms, the median age at diagnosis is 41 years, with males being significantly older at diagnosis than females. Prevalence rates ranged from 0,02% in 1994 to 1,2% in 2017. The average annual incidence was 87,7/100.000/year, increasing from 49,9/100.000/year (1994) to 96,21/100,000/year (2017). Athetosis is a neurological movement disorder characterized by slow, involuntary, and writhing body movements, especially of the limbs. It is often associated with abnormalities in the basal ganglia, a group of structures deep in the brain that are responsible for motor control. Athetosis is a subset of dyskinetic movement disorders and can occur as a primary condition or as a symptom of an underlying neurological condition. The term "athetosis" comes from the Greek words "a" meaning "without" and "thesis" meaning "position". It describes the inability to maintain a stable body posture due to the fluctuating and continuous movements observed in individuals with this condition. Athetosis can affect individuals of all ages, from infants to adults, and can have a significant impact on their motor function, coordination, and overall quality of life. In this case report, a woman under her family's care was reported with complaints of repeated twisting movements in her arms and legs which appeared suddenly and did not go away. The results of previous research show that cases dystonia are referred to neurosurgery for the management of dystonia, with modern neurosurgical management including pallidotomy, rhizotomy, and deep brain stimulation (Claassen et al., 2023; McEvoy et al., 2023). Meanwhile, there are several studies that show that this case often occurs in children (Kuzenkova et al., 2023). This research is different from previous research, this research took the case of Dystonia and athetosis in ICH of 80 years old female patient. This research contributes to increasing knowledge about dystonia and athetosis in ICH.

RESEARCH METHOD

This research is a qualitative research type with a case study approach (Sugiyono, 2020). The subject of this research is an 80 years-old woman. Research data uses primary and secondary data. Primary data came from examination results and secondary data was obtained from medical records, books and journal literature regarding this research. The data analysis technique uses qualitative data analysis techniques by concluding the results of the examination which are supported by objective data and subjective data which are supported by objective data and subjective data using literature reviews and book sources. (Karsadi, 2022; Nursapia, 2020).

RESULTS AND DISCUSSIONS

An 80-year-old woman came with her family to the emergency room at RAA Soewondo Regional Hospital, Pati with complaints of continuous movement of her right arm and leg. The movements carried out included rotating the arms and legs repeatedly without stopping. Complaints have been felt since two days of SMRS and appeared suddenly. According to the family information, the complaint was not better so the patient was taken to the emergency room. According to the patient's family, she did not consciously make such movements, and even though she was asked to stop, the patient did not stop. The movements stop only when the patient is sleeping. History of other diseases, trauma, and medication was denied, the patient had a history of high blood pressure (Di Biase et al., 2022; Dressler et al., 2022).

During a physical examination, continuous voluntary movements and twisting of the right arm and leg were found. Neurological deficits were also found in nerve VII, namely; eyebrows, nasolabial sulcus, and corners of the mouth that deviate to the left. Apart from that, there are also neurological deficits in nerve XII, namely; tongue deviation to the left side and tongue deviation during contraction to the right side. On motor examination, it was found to be 5-5-5-5/5-5-5-5, the physiological and pathological reflexes of the right limb were difficult to assess. For the left limb, it was within normal limits (Albanese et al., 2019; Frucht et al., 2021).

Supporting examinations are carried out in the form of laboratory examinations and radiological examinations. Laboratory results showed leukocytosis (11,8x10 ^ 3/ul), high neutrophil count (83,10), low lymphocytes (8,90%), and low eosinophils (1,50%). Obtained from the X-ray results of the chest, it showed cardiomegaly (RVH), suspected right lung tumor and atelectasis. A CT-Scan of the head without contrast was also carried out with the impression of bleeding in the left thalamus with minimal volume.

After the examination was completed, treatment was carried out in-patient and given infusion treatment asering 20 tpm, citicoline 2x1 tab, lansoprazole 1x1 tab, piracetam 3x1 tab, tranexamic acid 3x250 tab, omeprazole injection 1x1 ampoule, haloperidol 1x1mg tab, THP 1x2mg tab, syringe pump nicardipine 0.5mcq, and clonazepam 2x1mg tab.

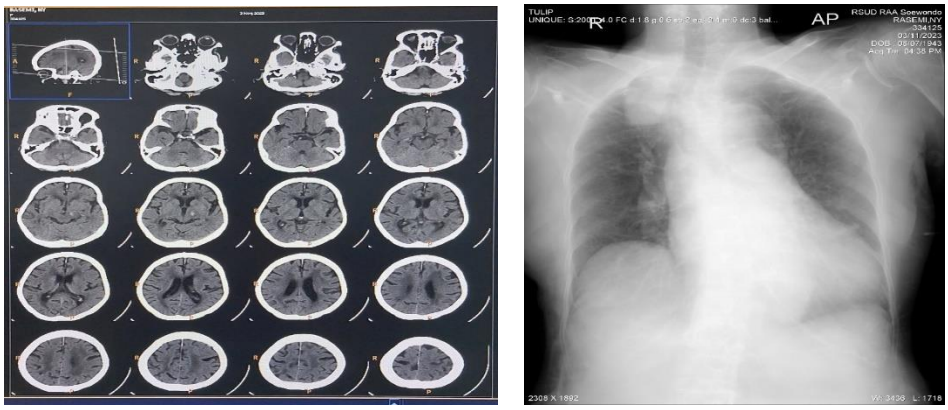

Figure 1. Supporting examination non-contrast head CT-Scan and AP chest x-ray

Discussion

In this paper, the author has presented an approach by collecting several sources to find out and discuss epidemiology, common causes, signs and symptoms, diagnosis, examination, treatment and complications that can occur. Dystonia is defined as "a movement disorder characterized by persistent or intermittent muscle contractions that cause abnormal, often repetitive movements, postures, or both. Dystonic movements are usually patterned, twisting, and may be shaking. It is often initiated or worsened by voluntary action and is associated with excessive muscle activation (Grütz & Klein, 2021).

The final definition of dystonia (proposed by consensus of the Movement Disorder Society expert members) is articulated in the following three sub-definitions dystonia is a movement disorder characterized by persistent or intermittent muscle contractions that cause abnormal and frequently repetitive movements, postures, or both, dystonic movements are usually patterned, twisting, and may be shaking and dystonia is often initiated or worsened by voluntary actions and is associated with excessive muscle activation (di Biase et al., 2022; Ercoli et al., 2021; Listik et al., 2023).

Body parts that experience dystonia usually remain mobile and the movements are often slow and suddenly fast. The most common forms of dystonia in adults are focal, affecting the neck (cervical dystonia), the eyes (blepharospasm), or task-related (e.g., *writer's cramp*). However, dystonia can also present with a generalized pattern, which is more common in childhood. Dystonia is a subcategory of hyperkinetic movement disorders in which movements appear excessive or irregular. Other subtypes include chorea, myoclonus, tics, and tremors (Albanese et al., 2011; Ercoli et al., 2021; Sadnicka et al., 2023).

Athetosis is a disorder of tortuous, slow, irregular, and involuntary movements that affects the distal extremities. Although the word is less commonly used, the term is still used to describe distal extremity dystonia in cerebral palsy as well as writhing movements of the fingers and/or toes due to loss of proprioception (pseudoathetosis). Athetosis usually occurs due to injury to the basal ganglia. In children, injuries often occur in the neonatal period, and many forms of cerebral palsy are athetotic. In adults, athetosis can occur in patients who have suffered a stroke. Sometimes, athetosis is seen as a long-term effect of cerebrovascular stroke (athetosis posthemiplegic). Pseudoathetosis may also be found in patients with severe peripheral neuropathy with loss of proprioception. Pseudoathetosis is not associated with functional disorders and does not require specific treatment because athetosis usually does not respond well to pharmacological treatment. Athethosis is part of extremity dystonia or limb dystonia (Albanese et al., 2013; Corp et al., 2019; Kilic-Berkmen et al., 2021; Lenka & Jankovic, 2021).

Dystonia is a very rare disease. This condition is experienced by 1% of the population worldwide, with more women than men suffering from it. Dystonia is a very rare case presenting to the Emergency Department (ED), comprising approximately less than 1% of all visits. In this case, dystonia with athetosis was suffered by an 80 year old woman who complained of continuous movement of the right arm and leg. The movements are such rotating the arms and legs repeatedly without stopping and appear suddenly since 2 days of SMRS (Dressler et al., 2021; Shetty et al., 2019).

Dystonia is a disorder heterogeneous in its clinical presentation and difficult to diagnose. Over time, several propositions for the classification of dystonia have been made. Nowadays, the 2013 consensus statement by the Movement Disorder Society (MDS) regarding the classification of dystonia has been effectively applied. However, these guidelines allow for leeway in clinical judgment, sometimes resulting in differing opinions or conclusions.[11] The most common area was the shoulder (13,4%), followed by hand dystonia (2,6%), upper face (2,1%), upper arm (1,2%), larynx (1,1%), lower face (0,7%), torso (0,7%), jaw, and tongue (0,3%).

The clinical diagnosis of adult dystonia is generally based on phenomenology. Most cases are idiopathic in origin, so no further testing is required to confirm the diagnosis. However, in a small proportion of patients, dystonia is caused by treatable congenital or genetic diseases (e.g. medication-induced dystonia, Wilson's disease, dopa-responsive dystonia), so prompt and accurate diagnosis is essential in these cases.

Axis 1: Clinical Characteristics of Dystonia

Age of onset can be further divided into childhood (0–2 years), childhood (3–12 years), adolescence (13–20 years), early adulthood (21–40 years), and late adulthood (>40 years) is most common).This is important, as there are different age distributions associated with subtypes different dystonias. Dystonia that begins under 20 years of age generally begins focally in the distal

extremities and is more likely to spread to other parts of the body. Dystonia that begins over the age of 20 is usually focal (most often affecting the neck, eyes, or limbs) or segmental (generally spreading craniocervically). Additionally, dystonia Late onset (>40 years) tends to occur earlier in the upper extremities and neck (cervical), compared to the face, jaw, and/or tongue (cranial). Dystonia cervical cancer generally appears in the fourth and fifth decades (mean age: 41,7 years), while dystonia spasmodic (laryngeal dystonia) tends to occur a little later (mean age of onset: 50,1 years). Rather, dystonia cranial consist of blepharospasm or oromandibular dystonia or a combination of both (Meige syndrome) occurs later in the sixth and seventh decades. Late - onset focal extremity dystonia occurs earlier, in the fourth decade.

Body distribution is determined based on the affected body region: (1) focal, if only one body region is affected; (2) segmental, if two or more adjacent body segments are affected; (3) multifocal, if at least two nonadjacent body parts (such as hands and feet) are affected; and (4) generalized, if the main torso and at least two other locations are affected. Hemi- dystonia , where there is combined unilateral upper and lower extremity involvement, is considered separately, and is often associated with dystonia acquired from contralateral brain lesions.

Temporal patterns were divided into four different categories: (1) persistent, (2) action-specific/task-specific, (3) diurnal fluctuations, and (4) paroxysmal. In a persistent pattern, dystonia is generally stable throughout the day (although it can be made worse by stress or other aggravating circumstances). Action-specific dystonia is a form of dystonia that occurs only when certain activities are performed. In the upper extremities, this usually involves fine motor activities that require precision, such as writing, typing, or playing a musical instrument. In the lower extremities, precision tasks can be performed (such as dancing/ballet), while endurance tasks, such as running or cycling are more frequently performed. Task-specific dystonia may involve other tasks, or even occur at rest over time. Diurnal fluctuations in severity dystonia may be present, especially in dopa-responsive dystonia occurring in childhood. However, many people find that the dystonia worsens over time, possibly related to increased fatigue. Lastly, dystonia paroxysms occur suddenly and in isolation, often associated with a specific trigger (movement, exercise, eating certain foods, physical/emotional stress), followed by the return of the underlying neurological condition.

Dystonia Paroxysms are very rare after these events. 18 years of age, unless there is an underlying lesion in the central nervous system. Occasionally, dystonic movements may occur in conjunction with other hyperkinetic phenomena, such as chorea or myoclonus. Therefore, the term paroxysmal "dyskinesia" is also used. Persistence of hyperkinetic movements after cessation of the trigger differentiates dystonia paroxysms of other forms of dystonia. In contrast, in task or action specific dystonia, the dystonia appears only when performing the task that prompted the dystonia to occur.

The presence or absence of non-dystonic clinical features is important to identify because if non-dystonic neurological features are present. This represents a much less common scenario and helps sharpen the differential diagnosis. Dystonia can be the only abnormal movement phenotype present (other than associated tremor), indicating isolated dystonia, or it can be combined with other movement disorders. In combined dystonia, there are other movement disorders besides dystonia, most commonly parkinsonism, myoclonus, or ataxia.

Axis 2: Etiology of Dystonia

Identifying the possible etiology for a particular form of dystonia can help determine the extent of testing needed and assist in selecting treatment. A history of intellectual or motor delays in childhood-onset dystonia helps guide the extent to which imaging and genetic testing may be useful. Signs of additional movement disorders, cognitive dysfunction, or other complex neurologic features in early or late adult dystonia also help guide the clinician in selecting investigations.

Adult-Onset Idiopathic Focal/Segmental Isolated Dystonia

Dystonia focal idiopathic onset in adults occurs less frequently, segmental dystonia is the most common form of dystonia overall and is not associated with features suggestive of a specific cause. This form of dystonia tends to be persistent and occurs less frequently with certain tasks. Spontaneous remission is rare in the first 5 years after disease onset. However, it often relapses. [20]

Dystonia Focal/segmental attacks in adults most commonly affect the cervical region. It often begins in middle age (30s-50s), often initially accompanied by neck pain, followed by the development of abnormal neck posture or jerky head tremors. Abnormal postures can include twisting or twisting of the neck, flexion, extension, lateral flexion, or a combination of these postures. In some cases, dystonic tremor of the head or hands is seen, which may resemble essential tremor. Sensory, which involves gentle touching of the head or neck, is often beneficial although this may diminish over time. Pain is a frequent and early presentation, associated with persistent abnormal posture caused by asymmetric muscle contractions, or rapidly alternating agonist/antagonist muscle activation in dystonic tremor, which causes focal muscle spasms in overused muscles and is associated with decreased quality of life. Patients may report a family history of dystonia and tremor in 14% and 29% of cases, respectively, suggesting the presence of polygenic genetics similar to essential tremor.

Dystonia cranial involves abnormal contractions of the muscles of the face and head. Subtypes include involvement of the eyelids (blepharospasm), lower face (commonly combined with eyelid spasms in *Meige syndrome,* or hemifacial spasms), or jaw or tongue (oromandibular dystonia), either separately or in combination. Local spread to the cranial area tends to occur commonly for months or years after the initial attack, as does extracranial spread to the neck and upper extremities with advancing age. Blepharospasm is the most common form. Initially, affected individuals show mild symptoms of increased bilateral eyelid blinking, sometimes triggered by exposure to bright light. If left untreated, blinking frequency will increase and may progress to longer seizures, which can limit vision, and approximately 12 to 36% may experience functional blindness. Simultaneous lower facial spasms suggest dystonia segmental craniocervical, called Meige syndrome. Sensory tricks include lightly touching the eyelids, applying pressure to the upper part of the orbit or canthus or rubbing the eyes.

Oromandibular dystonia includes dystonic involvement of various muscles of the jaw and mouth. This can occur with involuntary jaw opening, closing, lateral deviation, protrusion, or retraction movements, with or without jaw tremor. Such dystonia cervical, uncontrolled muscle contractions often lead to overuse of the muscles, resulting in jaw tension, pain, and spasms. These can also appear as bruxism. Activation of these muscles through speaking and chewing usually results in worsening of symptoms. However, there are several rare cases of paradoxical improvement when speaking or with certain tongue positions. Sensory tricks may include pressure on the lips or teeth, touching the tongue to the hard palate, chewing gum, or placing an object, such as a tongue depressor or toothpick between the teeth or in the cheek. Oral prosthetics have been used with some success. Tongue dystonia is a rare oromandibular dystonia that involves protrusion or curling of the tongue and can also cause drooling. Lingual dystonia most often occurs with speaking, and can cause problems with speaking, eating, and swallowing.

Dystonia (or spasmodic dysphonia) affects the vocal cords, causing difficulty speaking or singing. It is divided into two main forms, involving the adductor muscles (characterized by tense, strangled, and harsh speech) or the abductor muscles (causing breathless speech, worse with voiceless consonants). Spasmodic dysphonia is more common in women, and the adductor type is more common associated dystonic voice tremor occurs in 30% of cases. A useful clinical clue is that innate vocalizations (e.g., laughing, crying, or whispering) tend to be unaffected. Vocal difficulties may worsen with stress, when talking on the telephone, or when trying to project a voice, for example when speaking in public.

Systonia is rare in cases occurring in adults and is often caused by specific tasks. The most common upper extremity dystonia is writer's cramp, a task-specific dystonia that affects the hand, forearm, or upper arm when writing. Writer's cramp is more common in male, generally occurs in middle age, especially the fourth decade, and may lose its task specificity and involve tasks of the other hand or even spread to the contralateral hand as time goes by. Focal hand dystonia musicians generally engage in contraction of the fingers (often finger flexion) or wrist (less commonly) when playing a musical instrument. Musicians can also suffer from embouchure dystonia, a task-specific oromandibular dystonia, which affects the way the mouth or tongue plays woodwind or flute instruments. Such writer's cramp, dystonia musicianship is more common in men, and professional musicians, often occurring during the peak of their performance careers. The precipitating factors are overexertion or other hand injuries, as well as peripheral nerve entrapment in the motor distribution of the affected fingers, especially ulnar nerve entrapment, with the most common pattern being dystonic flexion of the ring and little fingers.

Lower extremity dystonia often involves running, but can also affect other activities, such as cycling or walking over certain terrain. Abnormal posture mostly involves plantar flexion, but may also involve proximal or distal muscles. Lower extremity dystonia that occurs in adults when isolated (i.e., not in the presence of PD) often begins with specific actions on the lower extremities, but often progresses to affect daily gait. Additionally, task-specific dystonia has also been reported in several different sports.

It appears that botulinum toxin (BoNT) treatment may have beneficial effects on some NMS, but reported results are conflicting. BoNT remains the mainstay treatment for dystonia focal. Its efficacy has been proven time in numerous studies and trials, with long-term follow-up also demonstrating continued safety and efficacy beyond two decades of treatment.[31,32] There are several different botulinum toxin formulations available today. A recent review concluded that, besides antiepileptics, for most pharmacotherapies nontoxin, evidence of its effectiveness based on research is still limited. Moreover, the genes of dystonia sufferers also influence treatment. In recent research, many new genes have been found in dystonia sufferers. The experimental pharmacotherapies mentioned are dipraglurant, this drug modulates metabolic glutamate receptor type 5 (mGluR5), which is involved in the development of dystonia in the DYT1 rodent model, and sodium oxybate, studied in alcohol-responsive spasmodic dysphonia patients.

Relevant generic therapy for all forms of dystonia paroxysms involve avoiding the trigger. In paroxysmal dyskinesia, low doses of carbamazepine (50-200 mg/day) can be very effective. Other antiseizure medications that may be effective in *paroxysmal kinesigenic dystonia* include oxcarbazepine, phenytoin, and lacosamide (50-100 mg/day), with possible benefits reported when given with valproic acid, lamotrigine, levetiracetam, or topiramate. In paroxysmal nonkinesigenic dyskinesia, treatment is less effective than PKD and includes low-dose benzodiazepines, gabapentin, and levetiracetam. The mainstay of treatment involves dietary glucose modification with a ketogenic diet, with concomitant L- carnitine, while triheptanoin (an odd-chain fatty acid) produces dramatic effects, reduction in PED attacks in an open-label trial. Data regarding the partial benefit of using levodopa, trihexyphenidyl, and benzodiazepines in PED are limited. There is little evidence to support surgical management of paroxysmal dystonia/dyskinesia.

Deep brain stimulation (DBS) is an effective advanced treatment option for dystonia refractory to treatment. Until today, the Globus Pallidus interna (GPi) has been the most frequently targeted area in dystonia patients. Its efficacy in dystonia focal, segmental, and generalized have been studied extensively in both short-term and long-term studies.

In general, the response of dystonia patients to DBS is variable and often difficult to predict. Approximately 10-25% of patients undergoing DBS can be considered nonresponders and experience improvement of less than 25%. Technical factors, such as electrode location play an important role in this but only explain part of the reduced response.[39]

For some patients, dystonic symptoms are not sufficiently controlled by DBS alone. Continuation of previous/additional therapy may be necessary. A long-term management study with a small population of cervical dystonia patients demonstrated optimal symptom control with at least one form of adjunctive therapy (BoNT and/or oral pharmacotherapy). Continuing additional therapy should be considered in patients who have not achieved satisfactory symptomatic improvement with sigle DBS treatment. Complications of dystonia can cause permanent bone deformities, contractures, and functional impairment.

CONCLUSION

Dystonia is a clinical diagnosis that is made based on history, observation and direct examination. Treatment depends on the patient's age, diagnosis, and distribution of dystonia, and is individualized, consisting of rehabilitation, botulinum toxin injections, oral pharmacotherapy, and DBS. Combination therapy can also be carried out pharmacotherapy and DBS. However, the greatest progress has been in the field of dystonia genetics, with many new genes discovered over the last 20 years. Analyzing the natural progression of each dystonia causing mutation, along with its response to treatment, can aid in identifying the most effective treatment for individuals with specific gene mutations. In this patient's case, the dystonia stemmed from intracerebral hemorrhage (ICH) in the basal ganglia, which also exhibited atheosis as part of the limb dystonia. The limitation of this research is that this research uses field cases with case handling according to health service standards. Future research should be able to carry out a more in-depth study using quantitative research to determine the relationship between dystonia stemmed from intracerebral hemorrhage (ICH).

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