Severity of Antipsychotic-Induced Cervical Dystonia Assessed by the Algorithm-Based Rating System

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Abstract

Background: The severity of antipsychotic-induced cervical dystonia has traditionally been evaluated visually. However, recent advances in information technology made quantification possible in this field through the introduction of engineering methodologies like machine learning.

Methods: This study was conducted from June 2021 to March 2023. Psychiatrists rated the severity of cervical dystonia into 4 levels (0: none, 1: minimal, 2: mild, and 3: moderate) for 101 videoclips, recorded from 87 psychiatric patients receiving antipsychotics. The Face Mesh function of the open-source framework MediaPipe was employed to calculate the tilt angles of anterocollis or retrocollis, laterocollis, and torticollis. These were calculated to examine the range of tilt angles for the 4 levels of severity of the different types of cervical dystonia.

Results: The tilt angles calculated using Face Mesh for each level of dystonia were $0° \leq \theta < 6°$ for none, $6° \leq \theta < 11°$ for minimal, $11° \leq \theta < 25°$ for mild, and $25° \leq \theta$ for moderate laterocollis; $0° \leq \theta < 11°$ for none, $11° \leq \theta < 18°$ for minimal, $18° \leq \theta < 25°$ for mild, and $25° \leq \theta$ for moderate anterocollis or retrocollis; and $0° \leq \theta < 9°$ for none, $9° \leq \theta < 17°$ for minimal, $17° \leq \theta < 32°$ for mild, and $32° \leq \theta$ for moderate torticollis.

Conclusion: While further validation with new cases is needed, the range of tilt angles in this study could provide a standard for future artificial intelligence devices for cervical dystonia.

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Advances in pharmacologic, physiological, and biological techniques have enabled the severity of various medical symptoms to be accurately assessed by laboratory and imaging findings. Assessing the severity of motor and behavioral abnormalities by these techniques has been difficult, so psychiatrists and neurologists have relied on visual impression and experience to evaluate these symptoms. However, recent advances in information technology and computer science have made quantification possible in this field, with the introduction of engineering methodologies such as machine learning.

Dystonia is a movement disorder characterized by sustained or intermittent muscle contractions causing abnormal, often repetitive, movements, postures, or both.[1] The term is used to describe an uncontrollable muscle spasm that becomes evident as a contraction of the flexor and extensor muscles, leading to an abnormal position. Symptoms include tongue protrusion, torticollis, retrocollis, trismus, oculogyric crisis, and pleurothotonus (eg, Pisa syndrome).[2] Of these, cervical dystonia is sometimes observed in psychiatric patients receiving antipsychotics. The unexpected bending of the neck, unsightly cervical appearance, and pain from pulled muscles often lead to the discontinuation of antipsychotic treatment, resulting in the relapse of psychosis. Therefore, detecting these minimal or mild symptoms accurately and treating them early may contribute to the prevention of nonadherence to antipsychotic treatment in the acute phase of antipsychotic treatment. Moreover, given that acute dystonia can be quickly reversed by anticholinergics or antipsychotic modification regardless of its severity, a precise assessment of dystonia in the chronic form may also be important for detecting the subtle change in the severity of tardive dystonia persistently observed in psychiatric patients over time.

The severity of cervical dystonia is usually assessed by the established 4-grade rating scale. However, the criteria for the boundaries differentiating the scores are ambiguous, and what levels are considered mild, moderate, or severe is largely left to the clinical impression of each physician. Evaluation by direct visual

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Clinical Points

- We developed an algorithm-based artificial intelligence (AI) rating system to assess the severity of antipsychotic-induced cervical dystonia, in which high reliability was confirmed with evaluations by trained psychiatrists.
- This AI-based application has the potential to quantify the clinical assessment of antipsychotic-induced dystonia by providing consistent and accurate ratings anytime, anywhere, without the need for a trained psychiatrist.

observation, without measuring the deviation angle, will seemingly inevitably result in variations.

Rater reliability on standardized rating scales such as the Drug-Induced Extrapyramidal Symptoms Scale (DIEPSS)[2–6] has been reported to be excellent among trained individuals, but achieving high reliability requires considerable training time. This artificial intelligence (AI)–based application has the potential to quantify the clinical assessment of antipsychotic-induced dystonia by providing consistent and accurate ratings anytime, anywhere, without the need for a trained psychiatrist.

This study investigated the concrete ranges of deviation angles for each severity level in psychiatric patients showing antipsychotic-induced cervical dystonia. It compared the rater consensus evaluation data and the deviation angle data using AI-based deviation angle measurement technology. We propose a global standard for evaluating the severity of abnormal deviation in cervical dystonia for the development of AI devices.

METHODS

Ethical Considerations

This study was conducted from June 2021 to March 2023, after the approval of the Ethics Committee of Nagoya University Graduate School of Medicine, Nagoya, Japan, following the committee's guidelines and regulations. All study procedures were conducted in accordance with the Declaration of Helsinki. Video recordings of psychiatric patients showing a variety of facial extrapyramidal symptoms of various severities were performed after obtaining the approval of the ethical committees of the attending individual institutions or hospitals. Written informed consent was obtained from all participants.

Cervical Dystonia Data Extraction From the Videoclip Library

The videoclip library of the DIEPSS[2–6] contains clips of various types of antipsychotic-induced extrapyramidal symptoms (gait, bradykinesia, sialorrhea, muscle rigidity, tremor, akathisia, dystonia, and dyskinesia). It has been used in DIEPSS training workshops for psychiatrists attending clinical trials. The consensus severity data assessed by trained psychiatrists are also stored with the corresponding videoclips. The data used in this study are based solely on this videoclip library recorded through December 2021. A total of 101 videoclips were extracted from this library. The distribution of subtypes of cervical dystonia is as follows: no cervical dystonia (n = 32), laterocollis (lateral bending) (n = 37), anterocollis (forward bending) (n = 17), retrocollis (backward bending) (n = 9), and torticollis (rotation) (n = 6).

Calculation of the Tilt Angle Using MediaPipe Face Mesh

The angle of cervical deviation was calculated for these 101 videoclips based on the deviation angle of the AI-based face. The range of angle for 4 severity levels (0: none, 1: minimal, 2: mild, and 3: moderate) was determined using the trained psychiatrist consensus scores in the DIEPSS videoclip library. Face Mesh, a solution in an open-source framework called MediaPipe, was used to calculate the tilt angle of the neck. Face Mesh is an analysis system that uses machine learning to detect 468 facial landmark points from the recorded videoclips. Figure 1 shows an example of the visualization of landmarks calculated by Face Mesh. Face Mesh can obtain the xyz coordinate data of each landmark point chronologically in each frame. The x-, y-, and z-axis values refer to the estimated distance from the camera and the horizontal and vertical coordinates in the frame, respectively. Furthermore, using the xyz coordinate data at these 468 landmark points, the roll, pitch, and yaw angles can be calculated, respectively. The roll, pitch, and yaw angles represent rotation angles around the x-, y-, and z-axes and correspond to the roll angle for the lateral bending angle, the pitch angle for the forward or backward bending angle, and the yaw angle for the rotation angle in dystonia symptoms, respectively.

Figure 2 shows the schematic imaging of the rotation direction by the type of cervical dystonia. As shown in this figure, the roll angle corresponds to the deviation of the laterocollis, the pitch angles correspond to the deviation of the retrocollis or anterocollis, and the yaw angle shows the deviation of the torticollis. The cervical tilt angles corresponding to individual posture types of cervical dystonia can be quantitatively calculated using Face Mesh from the videoclip data. We proposed using the technology to create (1) a range of tilt angles corresponding to the severity levels by the individual posture types of cervical dystonia and (2) an algorithm to evaluate the overall severity of a case video based on the quantitative angle information.

Figure 1.
An Example of the Landmark Points Obtained by Face Mesh

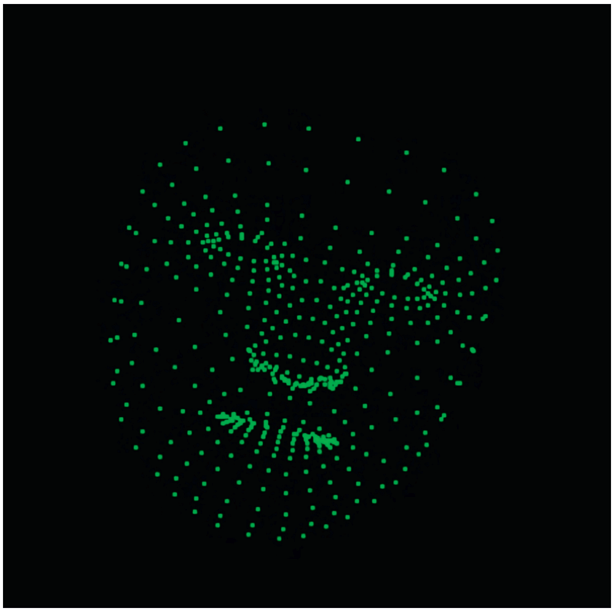
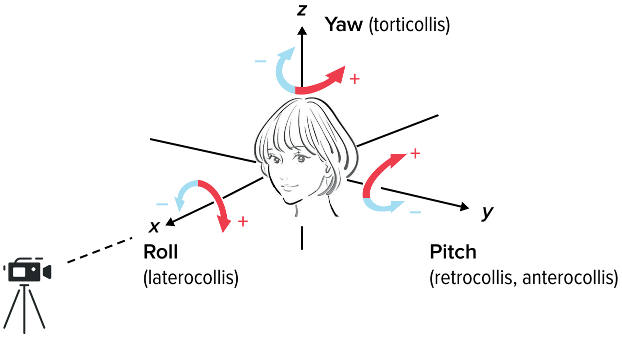


Figure 2.
Schematic Imaging of the Rotation Direction by the Type of Cervical Dystonia



Method of Determining the Range of Neck Tilt Angles for Individual Severities

We followed this method to develop an angle table defining the range of tilt angles corresponding to the 4 severity levels (0, 1, 2, and 3) in the 4 types of cervical dystonic postures.

1. The consensus severity (0, 1, 2, and 3) evaluated by the trained psychiatrists for 4 types of cervical dystonia in all 101 videoclips was defined as the gold standard severity (teacher labels) for each videoclip.
2. The tentative angle table showing the ranges of tilt angle for each severity level (0, 1, 2, and 3) in the 4 types of cervical dystonia was determined based on the empirical angle criteria by the trained psychiatrists.
3. Based on the tentative angle table, the severity of the 101 video cases was evaluated using the rules described in "Algorithm to Determine the Tilt Angle of Cervical Dystonia." The revised tentative angle table was then created.
4. The severity scores of the revised tentative angle table were compared with the consensus severity score evaluated by the trained psychiatrists and the severity score obtained from the algorithm-based cervical dystonia rating system. The agreement rates with the revised tentative angle table were recalculated.
5. The angle threshold of the revised tentative angle table was adjusted to minimize the disagreement rate between the consensus severity score evaluated by the trained psychiatrists and the severity score obtained from the algorithm-based cervical dystonia rating system.
6. Steps 3–5 were performed repeatedly, and the tentative angle table with the highest agreement rate was adopted as the final angle table.

Algorithm to Determine the Tilt Angle of Cervical Dystonia

This study used the following algorithm to determine the cervical dystonia tilt angle using the quantitative time series data of neck tilt angle calculated from case videos.

1. The roll, pitch, and yaw angles were calculated for each frame of the video to determine the severity of dystonia (Figure 1).
2. Based on the values of roll, pitch, and yaw angles and the angle range criteria table, the severity (0, 1, 2, and 3) of laterocollis, anterocollis or retrocollis, and torticollis (rotation) was determined for each frame.
3. For the severity of cervical dystonia with deviations in overlapping directions (rotation and lateral tilt in the same patient) in each frame, the highest tilt deviation among the 4 types of cervical dystonia was adopted as its type of cervical dystonia, and the severity was determined based on the angle range criteria.
4. The 95th percentile of the deviation angle was calculated from all frames, and its tilt angle value was adopted for evaluating the severity.

In this algorithm, by using the criteria of tilt angle range for individual severities (Table 1), a severity of 0, 1, 2, or 3 was first assigned to each frame for the 4 types of cervical dystonia. When assessing the severity of cervical dystonia in the dystonia item of the DIEPSS, the severity is assessed using not the angle of the average tilt over the assessment time but the angle at the point of maximum tilt observed over the entire

Table 1.

Range of Tilt Angles for Individual Severity Levels Obtained From the Consensus Evaluation by the Trained Psychiatrists

	Severity 0	Severity 1	Severity 2	Severity 3
Laterocollis	$0^\circ \leq \theta < 6^\circ$	$6^\circ \leq \theta < 11^\circ$	$11^\circ \leq \theta < 25^\circ$	$25^\circ \leq \theta$
Anterocollis	$0^\circ \leq \theta < 11^\circ$	$11^\circ \leq \theta < 18^\circ$	$18^\circ \leq \theta < 25^\circ$	$25^\circ \leq \theta$
Retrocollis	$0^\circ \leq \theta < 11^\circ$	$11^\circ \leq \theta < 18^\circ$	$18^\circ \leq \theta < 25^\circ$	$25^\circ \leq \theta$
Torticollis	$0^\circ \leq \theta < 9^\circ$	$9^\circ \leq \theta < 17^\circ$	$17^\circ \leq \theta < 32^\circ$	$32^\circ \leq \theta$

assessment. Therefore, the 95th percentile of the severity of all frames was adopted for the overall severity during the entire image in this algorithm. Here, the 95th percentile is a statistic value that refers to the point located at 95% when all frames are sorted in decreasing order of severity, counting from the smallest to the largest. To obtain the severest symptomatic timing, the adoption of the 100th percentile (= maximum value) could be considered, but the 95th percentile was adopted to avoid the problem that it would be greatly affected by outliers.

RESULTS

Characteristics of the Video-Recorded Patients

The selected 101 videoclips from the DIEPSS videoclip library were recorded from 87 patients (38 men and 49 women). The participants recorded were Japanese psychiatric patients receiving first- and/or second-generation antipsychotics. Their mean age at the time of recording was 49.6 ± 16.4 years (range 18–80). They were diagnosed with schizophrenia (62 patients) or mood disorders (25 patients).

Range of Tilt Angles for Individual Severities by the Type of Cervical Dystonia

Table 1 shows the range of tilt angles for individual severities by the type of cervical dystonia according to the description in "Methods."

Severity Agreement Rate

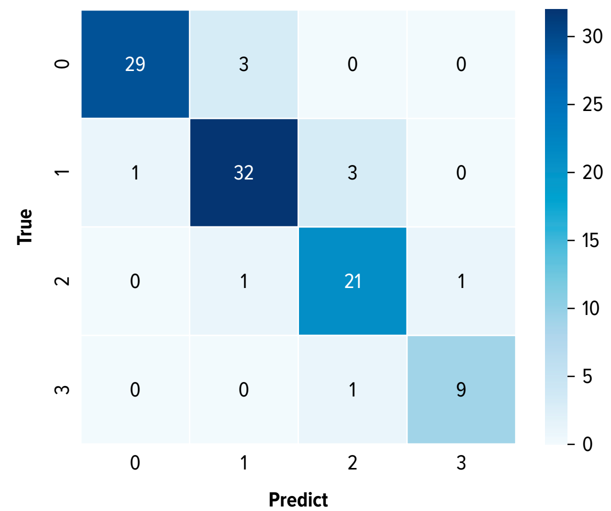
Figure 3 shows the crosstabulation of the concordance between the severity ratings based on the consensus by trained psychiatrists and those obtained from the algorithm-based cervical dystonia rating system. The simple complete agreement rate shown in Figure 3 is 0.901. The analysis of variance intraclass correlation coefficient (ANOVA ICC) value,[7] calculated using the SPSS version 28 program (IBM, Tokyo, Japan), was 0.950 (95% CI, 0.927–0.966).

A total of 10 cases did not show agreement between the severity ratings based on the consensus by trained psychiatrists and those obtained from the algorithm-based cervical dystonia rating system. The misclassification rates by severity (0, 1, 2, and 3) were 9.4% (3/32), 11.1% (4/36), 8.7% (2/23), and 10.0% (1/10) with $\chi^2 = 0.0872$, $df = 3$, and $P = .9933$. For types of dystonia (laterocollis, anterocollis, retrocollis, and torticollis), the rates were 8.1% (3/37), 11.8% (2/17), 11.1% (1/9), and 16.7% (1/6) with $\chi^2 = 0.4024$, $df = 3$, and $P = .9398$. The reasons for disagreement were as follows: data were located on the borderline differentiating between 2 severities (n = 6), the participant's posture was problematic (n = 3), and the point data could not be obtained with Face Mesh when the subject slumped down heavily (n = 1).

Table 2 shows the sensitivity and specificity results by the grade of severity.

Figure 3.

Crosstabulation Shows the Concordance Between the Severity Ratings Based on the Consensus by Trained Psychiatrists and Those Obtained From the Algorithm-Based Cervical Dystonia Rating System[a]



[a]"True" indicates severity ratings based on the consensus by trained psychiatrists; "predict" indicates severity ratings obtained from the algorithm-based cervical dystonia rating system.

DISCUSSION

Severity ratings in psychiatry have sometimes differed for the same event, depending on the evaluator's views and values. In addition, clearly separating borderline abnormalities using the empirical human eye has been sometimes difficult. However, recent advances are bringing AI evaluation systems into the world of psychiatric interviews to obtain stable evaluations anytime and anywhere. When using rating instruments based on AI technology, sufficient reliability must be established between the rater's evaluation and the rating

Table 2.
Sensitivity and Specificity Results by Grade of Severity

	Severity 0	Severity 1	Severity 2	Severity 3
Sensitivity	0.906	0.889	0.913	0.900
Specificity	0.986	0.938	0.949	0.989

application evaluation. In the present study, 4 levels of severity were differentiated clearly, with high reliability between the rater's evaluation and the evaluation by the rating application. The use of this application is expected to reinforce the human judgment that has been made visually until now. The algorithms presented here show the concrete ranges of tilt angle for individual severity levels. The fact that the evaluation is no longer divided according to differences in skill level and that medically accurate results could be obtained anytime and anywhere will be of significance.

Several instrumental techniques have been proposed to accurately rate the severity of movement disorders. These encompass tremors,[7,8] gait disturbances,[9] dyskinesia,[10] and akathisia.[11] These techniques are primarily developed for research purposes to collect precise, consistent data elucidating the pathophysiology of movement disorders. While the technical data and calculations for these devices are impressive, their practicality in clinical settings remains a matter of debate, especially when considering the cost and time required for evaluation. Some are not widely used in daily clinical practice due to disadvantages such as requiring expert knowledge for data analysis or discomfort in wearing the device.

Although psychiatrists have previously evaluated the severity of illness using integer values (such as 0, 1, 2, and 3), the newly developed algorithm-based evaluation system will also enable continuous scoring of severity using the level of decimal values and allow for more sensitive evaluation. For example, when the severity improves from 2.4 to 1.9, this would show an improvement of 0.5 points, whereas the same mild level (2) would remain by the current human evaluation. Indeed, the question of whether a change of 0.5 points is clinically meaningful is of critical importance. For instance, to detect mild dystonia in its earliest stages and prevent its subtle progression with each visit, assessing the disease's progression with decimal point accuracy is beneficial. Such nuanced changes might not be immediately evident to psychiatrists, but an accurate assessment would be critical to the patient's treatment. Nevertheless, it is also important to note that tardive movements can fluctuate between visits. This fluctuation may obscure the detection of small changes, suggesting that a single evaluation may not always provide a complete picture. Therefore, considering a trend or trajectory across several visits might be of more clinical significance.

Anatomically, the neck has been treated as one localized area for the evaluation of dystonia. Considering that different muscle groups cause different phenotypes of cervical dystonia, even when, for example, laterocollis and torticollis occur simultaneously, using this evaluation system could make it easy to know that multiple muscles are involved from the angle of deviation.

A number of limitations could be considered in the present study. First, since the measurement of this study focuses exclusively on the most severe abnormal position of cervical dystonia, it may not be applicable to cases with severe movement disorders that could interfere with the deviation measurement. Second, in this boundary value determination, the angle was determined in 1-degree increments. Of the 10 cases whose severity ratings did not match, 6 were identified as borderline severity. These disagreements may be improved by adjusting the boundary value in 0.1-degree increments or adjusting the percentile point used to determine the overall severity in the final determination. These differences may reflect effects such as Müller-Lyer illusion of the human eye; in fact, the human eye–judged scores for borderline severities may be incorrect in terms of the accuracy of the deviation angle. Third, the evaluation images were taken in an improper position in 3 cases. Since the algorithm developed here uses only the facial landmark points obtained by Face Mesh and does not have a function to compensate for the original postural tilt, the evaluation of these 3 cases was not consistent with the consensus assessment scores by the trained psychiatrists. Since cases of cervical dystonia may include other dystonia such as shoulder elevation, adding an evaluation function that includes shoulder elevation must be considered in the first step of algorithm development in the future. The future development of the algorithm must combine techniques for detecting posture information and consider adding a function to compensate for posture tilt. In 1 case, Face Mesh could not acquire point data when the participant suddenly turned to the side. This could be due to the influence of the shooting conditions. Since Face Mesh is a technology that inputs camera images, it will inevitably be affected by shooting conditions. This could be resolved by clarifying the shooting conditions. Furthermore, the assessment methodology developed in this study for cervical dystonia may also have significant implications for evaluating other forms of antipsychotic-induced movement disorders in the facial region, such as tremor (eg, rabbit syndrome) and dyskinesia, as assessed by the DIEPSS. A comprehensive assessment of these movement disorders is crucial, particularly in patients who present with multiple types of movement abnormalities, as it aids in making informed treatment decisions. Fourth, since the DIEPSS evaluates the severity of dystonia based on both the degree of neck tilt and its impact on quality of life (QOL), the tilt angle assessed in this study is only one factor. Given the varying impact of the tilt angle on QOL among patients, potential misclassification might arise in part from the intricate relationship between neck tilt and its effect on QOL.

In this study, the 4-grade severity criteria for assessing the level of antipsychotic-induced cervical dystonia were identified as specific ranges of deviation angles. Although the current assessment criteria specifically focus on patients with antipsychotic-induced cervical dystonia, the techniques presented in this study may also be applicable to a broader range of patients with other etiologic forms of cervical dystonia, including idiopathic, genetic (hereditary), and functional (psychogenic) types. While further validation is needed by adding newly recruited patients, these differentiation criteria could be used as a global standard for determining the severity of abnormal deviation in cervical dystonia for the development of AI devices.

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