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SHORT COMMUNICATION

Dystonic head tremor in paroxysmal dyskinesia in 17 dogs (2021–2023)

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Abstract

Background: Dystonia is a common component of the movement disorder paroxysmal dyskinesia (PD) in dogs. However, the incidence of dystonic head tremor (DHT) in these dogs has not previously been evaluated.

Methods: The medical records of dogs presenting with PD between 2021 and 2023 were retrospectively reviewed, and those with available video footage and the presence of a head tremor were selected for further analysis.

Results: Seventeen of the 39 (43.6%) dogs diagnosed with PD that had video footage available manifested DHT. Poodle or Poodle-cross was the most commonly affected breed (7/17). DHTs were described as fine irregular head tremors accompanied by cervical dystonia (17/17), truncal (11/17) or head (10/17) sway, shifting limb (10/17) or single limb (6/17) dystonia, freezing (8/17), ataxia (6/17), ptyalism (5/17), falling (5/17), kyphosis (4/17) and prayer posture (4/17). Neurological examination and advanced imaging, when available, were within normal limits.

Limitations: The limitations of the study include its retrospective nature, the lack of video recordings for all PD patients and the lack of electrophysiological evaluation of tremors and electroencephalography.

Conclusions: DHT exists in dogs with PD; it has characteristic features, and it should be considered in differential diagnoses for dogs with head tremors.

INTRODUCTION

Dystonia and tremor are movement disorders that can occur independently or coexist.[1] Dystonic tremor describes the tremor in a body part that is affected by dystonia and is predominantly a postural/kinetic focal tremor with irregular amplitudes and variable frequency (mainly less than 7 Hz) that usually does not occur during complete rest.[2] Dystonic tremor can appear in any part of the body affected by dystonia; however, is most commonly seen as dystonic head tremor (DHT) in humans with the movement disorder cervical dystonia.[3,4] Although dystonic tremor is difficult to distinguish from non-dystonic tremor,[1] when head tremor is accompanied by concurrent abnormal posture of the head and neck, a diagnosis of dystonic tremor is more likely.[2] The pathophysiology of dystonic tremor is unknown, but basal nuclei pathology has been postulated.[5]

In dogs, although dystonia is a common component of the movement disorder paroxysmal dyskinesia (PD), tremor has also been reported within its clinical spectrum.[6] More specifically, isolated head but also limb or generalised tremors have been reported briefly in paroxysmal non-kinesigenic dyskinesia in Toy Poodles,[7] Chinook dogs,[8] Labrador Retrievers[9] and Maltese dogs,[10] in paroxysmal gluten-sensitive dyskinesia in Border Terriers[11] and in paroxysmal exertional dyskinesia in Shetland sheepdogs[12] and Welsh Terriers.[13] In a recent retrospective study of 31 dogs with PD, head tremors, head bobbing and whole body or limb tremors were reported in 11%, 6% and 39% of the population, respectively.[14] The aim of this study was to identify the incidence of DHT in a population of dogs diagnosed with PD and describe its clinical features.

MATERIALS AND METHODS

This is an observational single-centre retrospective study conducted in a veterinary teaching hospital

between 1 January 2021 and 1 January 2023. Ethical approval was not required for this study; consent forms for the usage of medical records and video footage of each patient had been signed by all owners at the time of admission. Cases were recruited using electronic records. The search terms included PD. Inclusion criteria consisted of (a) complete medical records, (b) a clinical diagnosis of PD based on published criteria (dystonic signs, absence of autonomic signs other than gastrointestinal signs, absence of postictal signs, presence of consciousness, normal neurological examination between the episodes),[6] (c) availability of video footage and (d) presence of head tremor. Specifically for the autonomic signs, we did not exclude gastrointestinal signs (e.g., hypersalivation, vomiting) as they have been reported previously in dogs with presumptive PD.[11,13,15-17] Complete medical records consisted of minimum data, including signalment, presenting complaints, clinical and neurological findings and final diagnosis. If available, clinicopathological, magnetic resonance imaging (MRI) and cerebrospinal fluid (CSF) analysis findings were also collected. All clinical and neurological examinations and video assessments were performed by a board-certified neurologist or a neurology resident under the direct supervision of a board-certified neurologist. MRI used a high-field magnet (1.5 T Intera, Philips Healthcare) and included a minimum of transverse and sagittal plane T2-weighted, fluid attenuation inversion recovery and T1-weighted pre- and post-contrast (gadopentetate dimeglumine, 0.1 mmol/kg intravenous bolus) images. Descriptive statistical analysis was performed using standard statistical software (SPSS Statistics 26, IBM Corporation).

RESULTS

One hundred and four dogs with PD were admitted during the study period, of which only 39 had video footage available. Of these 39 dogs, 17 (43.6%) met the inclusion criterion of manifesting DHT. The breeds represented in the final sample were Poodle-crosses or Toy Poodles (7/17; 41.2%), Boston Terriers (2/17; 11.8%), Chihuahuas, French bulldogs, Husky dogs, Pomeranians, Shetland sheepdogs, Shih Tzus, Maltese-crosses and crossbreeds (1/17; 5.9% each). Of the 17 dogs, 12 (70.6%) were male and five (29.4%) were female; 12 (70.6%) dogs were neutered and five (29.4%) were entire. The median age at presentation was 5.2 years (range: 6 months–10.4 years; interquartile range [IQR]: 3.9 years). The median length of time between the first episode and presentation was 1 year (range: 1 month–10 years; IQR: 1.5 years). The median bodyweight was 9.8 kg (range: 1.7–18 kg; IQR: 7.8 kg). All dogs presented due to paroxysmal episodes. Anxiety was reported between the episodes in five of 17 (29.4%) dogs.

The semiology during an episode in dogs with PD included DHT manifested as fine irregular head tremor, all of which were accompanied by cervical dystonia. Cervical dystonia included head turn (7/17; 41.1%), head turn and tilt (3/17; 17.6%), head tilt (2/17; 11.8%) and neck extension upright (retrocollis), retrocollis and head turn, neck ventroflexion (anterocollis), anterocollis and head turn (1/17; 5.9% each). Other signs included truncal sway (titubation) (11/17; 64.7%), head sway (10/17; 58.8%), shifting limb dystonia (10/17; 58.8%), single-limb dystonia (6/17; 35.3%), freezing (8/17; 47.1%), kyphosis (4/17; 23.5%), prayer posture (4/17; 23.5%), falling (5/17; 29.4%), ataxia (6/17; 35.3%), vomiting after episode (2/17; 11.8%), lipsmacking (3/17; 17.6%) and lip dystonia ('showing teeth') (1/17; 5.9%) (Video 1). Posture during episodes included sternal recumbency (8/17; 47.1%), standing up (3/17; 17.6%), sitting on pelvic limbs and then falling on lateral recumbency (3/17; 17.6%), walking, sternal-to-lateral recumbency and sternal recumbency to walking (1/17; 5.9% each). No autonomic signs, with the exception of gastrointestinal signs, were observed. Ptyalism without foaming (5/17; 29.4%) was present. The mean duration of the paroxysmal episodes was 5 minutes (range: 2–27.5 minutes; IQR: 5.5 minutes). Clinical and neurological examinations were unremarkable in all dogs.

All dogs had complete blood counts and serum biochemistry that were unremarkable. MRI of the head was performed in eight of 17 (47%) dogs, and this revealed only incidental findings unrelated to PD in a few dogs: supracollicular fluid accumulation (2/17; 11.8%), Chiari-like malformation and syringomyelia (1/17; 5.9%) and ventricular asymmetry (1/17; 5.9%). In dogs with supracollicular fluid accumulation, the parenchymal compression was lower than 14%, and there was no compression of the cerebellum; therefore, they were considered of no clinical significance.[18] Cisternal cerebellomedullary CSF analysis was performed in four of 17 (23.5%) dogs and was within normal limits. Serology for anti-canine gliadin immunoglobulin G (IgG) and anti-canine transglutaminase-2 IgA was not performed for any of the dogs. All dogs were diagnosed with presumptive inherited paroxysmal non-kinesigenic dyskinesia.

No treatment was pursued in 10 of 17 (58.8%) dogs. Four dogs (23.5%) received levetiracetam (20–30 mg/kg orally every 8 hours), which did not alter the episode frequency. A gluten-free diet trial was tried in three of 17 (17.6%) dogs; two dogs had no change in episode frequency, while one dog had a reduction in episode frequency for 8 months and then an increase (one episode every 6 weeks). Medications prescribed by the referring veterinarian were discontinued due to no change in episode frequency (imepitoin, $n = 2$; phenobarbital, $n = 1$; levetiracetam, $n = 1$).

DISCUSSION

This is the first study to evaluate the manifestation of DHT in a population of dogs with PD and describe its clinical features. All dogs with DHT had a simultaneous involuntary abnormal posture of the neck and

head (e.g., head turn, head tilt) that could potentially resemble cervical dystonia as described in humans. The clinical features of DHT consisted of fine irregular head tremor that occurred during posture (standing up, sternal or lateral recumbency) and usually coexisted with trunk or limb dystonia. The incidence of DHT in the population of dogs in this study was 43.6%, although this might be inaccurate based on the fact that not all dogs presenting with PD dogs had video footage available. Poodle or Poodle-cross dogs were the most commonly affected breed, and a potential genetic background associated with the specific phenotype of PD, including tremors, could be speculated. In humans, DHT seems to be a frequent feature (58.3%) of the movement disorder cervical dystonia,[3] and the prevalence of dystonic tremor in dystonic syndromes varies from 11% to 87% across studies in humans.[4] DHT in humans manifests during action or while maintaining posture; it has a low frequency (<7 Hz), is rhythmic but occasionally irregular and can vary in amplitude or presence based on positioning.[4] It usually worsens when the patient voluntarily moves the affected body part against the major direction of pulling caused by dystonia.[5] Differential diagnoses of DHT in humans include essential tremor, Parkinsonian tremor, cerebellar (intentional) tremor and head-bobble doll syndrome.[19,20] In dogs, the clinical features of DHT appear to be similar to those in humans; however, electromyography during tremor could not be performed to identify its frequency due to its episodic nature. As essential head and Parkinsonian tremors have not been reported in dogs, differential diagnoses for DHT in dogs are limited to cerebellar (intentional) tremors, episodic non-intentional head tremor (idiopathic head tremor) and other non-tremor disorders, such as head myoclonus, peripheral nerve hyperexcitability or focal seizures.

Limitations of this study include its retrospective nature, lack of systematic recording of episode semiology and video recordings in all PD patients and lack of electrophysiological evaluation of tremors and electroencephalography.

In summary, the present study indicates that DHT exists in dogs and appears to occur frequently (43.6%) in this population. DHT should be considered as a clinical manifestation of dystonia within the spectrum of PD in dogs, and it should be distinguished from other types of isolated head tremors.

AUTHOR CONTRIBUTIONS

Conceptualisation, methodology, investigation, analysis and writing—original draft and review and editing: Theofanis Liatis. *Methodology, analysis, review, editing and supervision*: Steven De Decker.

CONFLICT OF INTEREST STATEMENT

The authors declare no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

No ethical approval was required for this study.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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