REVIEW ARTICLE

Dystonia in Performing Artists: Beyond Focal Hand Dystonia

Somdattaa Ray, Pramod K. Pal

ABSTRACT: Overuse of specific muscles in perfecting movements in performing arts makes an artist prone to many medical conditions. Musicians' hand dystonia is focal task-specific dystonia (FTSD) of hand among musicians that has been extensively studied. However, embouchure, lower limbs, and laryngeal muscles can also be affected among musicians. Embouchure dystonia (ED) refers to dystonia of the perioral and facial muscles that occurs in musicians while playing embouchure instruments. It is essential to identify ED since the dystonia might become persistent and non-task-specific if the musician continues to play the instrument. Task-specific dystonia of lower limbs among musicians has been exclusively reported among drummers. The diagnosis rests on electromyogram (EMG) of the involved muscles during the task. Singer's dystonia (SD) refers to task-specific laryngeal dystonia that occurs only while singing. The diagnosis of SD is based on laryngeal EMG and spectrographic analysis. Cortical hyperexcitability, loss of inhibition, and aberrant plasticity are central to the pathogenesis in both ED and musicians' hand dystonia. The pathophysiological studies in SD are limited. This review aims to discuss the lesser known dystonias among performing artists – ED, FTSD of lower limb, and SD.

RÉSUMÉ: Manifestations de la dystonie chez les artistes de la scène : aller au-delà de la dystonie focale de la main. Le fait d'utiliser excessivement des muscles spécifiques en vue de perfectionner certaines tâches peut rendre les artistes de la scène sujets à de nombreuses affections. À cet égard, la dystonie focale de la main chez les musiciens a été largement étudiée. Cela dit, tant les muscles du visage que ceux des membres inférieurs et ceux dits laryngés peuvent aussi être affectés chez les artistes de la scène. La « dystonie de l'embouchure » fait ainsi référence à un trouble des muscles péribuccaux et faciaux qui survient au moment de jouer des instruments qui possèdent une embouchure. Il est donc essentiel d'identifier ce type de dystonie en raison de la possibilité qu'elle devienne persistante et non reliée spécifiquement au fait de continuer à jouer d'un instrument. Des cas spécifiques de dystonie des membres inférieurs ont par ailleurs été signalés exclusivement chez des batteurs. Un diagnostic reposera sur un test d'électromyographie (EMG) portant sur les muscles sollicités au moment de jouer d'une batterie. La dystonie des chanteurs renvoie quant à elle à un trouble qui affecte les muscles laryngés et qui survient uniquement pendant qu'une personne chante. Dans ce cas, un diagnostic sera basé sur un test d'EMG de ces mêmes muscles et sur une analyse spectrographique. Mentionnons que l'hyperexcitabilité corticale, la perte d'inhibition ainsi qu'une plasticité cérébrale de nature aberrante sont des éléments centraux de la pathogénie des muscles du visage et de ceux des mains chez les musiciens. Les études physiopathologiques de la dystonie qui affecte les chanteurs sont toutefois limitées. Bref, cet article entend aborder les dystonies moins connues que l'on trouve chez les artistes de la scène (dystonie des muscles du visage, dystonie spécifique liée aux membres inférieurs, dystonie des chanteurs).

Keywords: Musician dystonia, Performing artists, Embouchure dystonia, Percussionist, Singer

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Introduction

Performing artist refers to an artist who is involved in a theatrical presentation that incorporates various art forms, such as dance, sculpture, and music. The subtlety of performances by artists speaks of the time dedicated to learning and perfecting the intricate movements in the performance. However, their performances may be debilitated by many medical conditions such as neuromuscular injury, tendinitis, and focal dystonia.[1]

Dystonia is the commonest movement disorder encountered in performers. It is characterized by sustained, involuntary, and repetitive contractions of muscles causing abnormal posturing or twisting of a body part.[2–5] Focal task-specific dystonia (FTSD) refers to continuous, involuntary muscle contractions of a single body part that manifests during a particular activity and results in abnormal posturing of the body part.[6]

Task-specific dystonias have been described among performing artists. The term "musicians' dystonia" refers to task-specific dystonia among musicians while playing musical instruments. Among performing artists, focal dystonia of hand has most commonly been described among the musicians. However, task-specific dystonia among musicians is not limited to hands alone. Musicians playing embouchure instruments and those playing lower limb percussion instruments and singers who extensively use their vocal cords are also at risk of developing FTSD of the overused muscles. These

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dystonias include embouchure dystonia (ED), lower limb drummers' dystonia, and singer's dystonia (SD). The literature is replete with the vast repertoire of knowledge of phenomenology, pathogenesis, and therapies in the field of musicians' hand dystonia, while knowledge in the rarer forms of dystonia in performing artists has been relatively limited. This review aims to highlight the lesser known FTSDs among performing artists –ED, lower limb drummers' dystonia, and SD. Although task-specific dystonia of lower limb has also been seen among dancers, this dystonia has not been discussed in view of paucity of literature. The first part of the review delineates the phenomenology and diagnosis of these dystonias, and the second part discusses the pathogenesis of ED and FTSD of lower limb. The possible pathogenetic mechanisms behind the decreased propensity of lower limbs to develop task-specific dystonia in musicians are also discussed.

Search Strategy

Search for relevant articles was carried out using PUBMED database on 18 July 2020, and the references were selected based on relevance to this topic. Using the terms "embouchure" and "dystonia," the search results showed 34 articles, of which 25 articles were selected.

Using the terms such as "percussionist," "drummer," and "dystonia," 21 articles were found out of which 5 articles were selected. Certain case series on FTSD of lower limb have included percussionist dystonia. To identify these articles, search terms such as "lower limb" and "dystonia" were used, showing 393 results out of which 3 articles were selected.

Search using the terms such as "singer" and "dystonia" yielded 78 results. Six articles were relevant to this review and were selected.

Clinical Features

Embouchure Dystonia (ED)

Embouchure refers to muscles of lips, tongue, jaw, and face that function harmoniously to modulate airflow into mouthpiece of an instrument.[7] ED refers to dystonia of the perioral and facial muscles that occurs in musicians while playing embouchure instruments.[8] Task-specific dystonia involving the oral muscles has been reported in prayer-induced lingual dystonia[9] and auctioneer's jaw.[10]

Epidemiology

The exact incidence of ED is not known. In a study conducted by Moura et al., of the 2233 musicians studied, 318 musicians were identified to have motor control disorders. Among them, 49 musicians (2.2%) were classified as having musicians' dystonia. Among 22 wind players, 16 reported ED (70%).[11] In another study of 119 musicians with musician dystonia (MD), ED was observed in 19% of the patients.[12] In a study of 585 brass musicians, 68% of musicians reported embouchure problems.[13] The age of onset of ED ranged from 16 to 66 years.[7,8,14–16] Among these individuals with ED, the age of starting the training ranged from 8 to 22 years.[8,14] Individuals with ED reported performance impairment 1 month to 27 years after the onset of symptoms.[7,14] Among musicians with ED, 70%–78% of the patients were males.[7,14]

Precipitating Factors

Unlike musicians' hand dystonia where a triggering factor such as ulnar, median, or posterior interossei neuropathy,[17,18] or recent change in the intensity or pattern of practice[16,17] has been documented, the association between the similar trigger factors and ED is weak. Patients have reported an increase in practice time or precision in practice prior to the onset of ED,[8] but the number of patients reporting this change is negligible.[14] In a study by Frucht, history of trauma to the oral cavity was reported by 2% of the patients.[14]

Phenomenology

Embouchure instruments have been classified into brass (French horn, trumpet, trombone, and tuba), single reed (clarinet and saxophone), double reed (oboe and bassoon), and flute.[7,8,14] Extremely high force of 50–100 pounds per square inch is used while playing high-register brass instruments that could be a cause for the abnormal distortions.[19]

Classification of Phenomenology

The phenomenology of ED has been classified into six phenotypes – embouchure tremor (ET), lip-pulling phenotype, lip-locking, Meige's syndrome, jaw dystonia, and tongue dystonia.[7,14] The lip tremor appears while performing the task, rather than assuming the position of playing the instrument.[8] Both upper and lower lips are involved, and the frequency of oscillation is very high. In the lip-pulling phenotype, lateral deviation of the lip or angle of mouth, and outward protrusion of one or both lips are seen. In the lip-locking phenomenology, lips involuntarily seal around the mouthpiece of the instrument resulting in airflow impairment.[8]

Jaw dystonia phenotype includes jaw tremor, jaw closure, and anterior or lateral movement of jaw.[8,14] The dystonia phenotype is dictated by the instrument that is played (Table 1). Register of an instrument refers to the pitch of the sound produced by the instrument. The pitch of the sound emanating from a low-register instrument is lower than that produced by a high-register instrument. Embouchure instruments are classified into low-register and high-register instruments. Low-register brass instruments' players such as trombone and tuba report lip-locking movements, whereas high-register brass instruments such as trumpet and brass horn show lip-pulling movements and ET. Tongue dystonia and jaw dystonia are most commonly seen in woodwind instrument players. The jaw plays an important role in playing reed instruments and hence jaw dystonia phenotype is commonly seen in these musicians.[7] Apart from instrument specificity, the dystonia is register-specific (i.e., dystonia develops while playing at a particular pitch), speed-specific as well as technique-specific.[20] Register and technique specificity were observed in 62.1%–71% and 41.3%–51% of the musicians with ED, respectively.[7,14] As the disease progresses, other registers eventually get affected.[14]

Spread of Dystonia in ED

ED has been reported to spread to activities such as speaking, eating,[7] and drinking from bottle with small aperture.[8,14,21] It is most commonly associated with jaw dystonia phenotype.[7,8,14] Spread of dystonia has also been observed among patients with ED who have

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Table 1: Phenomenology and epidemiology of ED

Author	Patients	Findings
Moura et al., 2017[11]	Musicians(n = 2233) were evaluated	Among wind players (n = 22), 16 patients had ED.
Termsarasab and Frucht, 2015[7]	Patients with embouchure dysfunction (n = 139)	109 patients had ED. Trumpet and French horn players had tremor. Jaw dystonia was observed in reed players. Lip-pulling dystonia was seen in flutists.
Frucht, 2016[20]	Videos of patients with ED were assessed (n = 65)	Evaluation of buzzing, using O zone, and playing with mouthpiece of the instrument only helped in identification of ED. ED showed technique (n = 1), register (n = 4), speed (n = 1), and instrument specificity (n = 1). Geste antagoniste is seen in 10% of patients with ED (n = 7).
Steinmetz et al., 2013[13]	Brass musicians (n = 585)	Embouchure disorders were observed in 58.6% of the musicians. Female musicians reported higher incidence of embouchure disorders including embouchure cramping. Brass musicians were 1.7–2.1 times more likely to report lip cramping following change in embouchure technique.
Altenmuller et al., 2012[24]	FTSD in 591 musicians compared to 2651 healthy musician students	ED was seen in 20.1% of the patients.
Schmidt et al., 2011[12]	MD (n = 116)	ED was seen in 19% of musicians with MD.
Frucht, 2009[14]	Musicians with ED (n = 89)	Lip-locking movements was seen in low-register brass instruments. Lip-pulling and ET were seen in high-register brass instruments. Woodwind instrument players showed tongue dystonia (in flute layers) and jaw dystonia. Dystonia while performing oral tasks was seen in 15% of the musicians with ED. Dystonia while speaking was seen in 38% of musicians with jaw, tongue dystonia, and Meige's phenotype. Technique specificity was seen in 51% and register specificity in 71% of the patients. Preexisting writer's cramp was seen in 6%. Sensory perception was normal over the embouchure.
Rosset Llobet et al., 2007[23]	Musicians with MD (n = 101)	ED was seen in 18 musicians. Musicians playing saxophone, clarinet, and flute demonstrated only FTSD of hand, whereas musicians playing trumpet, horn, and tuba had exclusive lip involvement. Involvement of both hand and embouchure was seen in flute and saxophone, in different individuals.
Ragothaman et al., 2007[25]	ED in Nadaswaram player (n = 1)	Lower lip dystonia was present. Patient later developed postural tremor of upper limb. He had family history of essential tremor.
Frucht et al., 1999[21]	French horn players (n = 2)	Lip-pulling dystonia was observed. In one patient, dystonia developed on drinking from soda bottle.
Morris et al., 2018[31]	Brass players with ED (n = 9), healthy brass players (n = 6)	Acoustic analysis revealed that musicians with ED had tremor of 5–8 Hz, sound instability, attack inaccuracy (impaired pitch initiation), and sound breaks.
Lee et al., 2014[30]	Professional musicians with ED (n = 7), healthy controls (n = 10)	Fundamental frequency showed higher fluctuation in all registers in musicians with ED. The fluctuation ranged between 5 and 25 Hz.
Lee et al., 2016[32]	Professional musicians with ET (n = 7), and healthy professional and nonprofessional musicians (n = 7)	ET has a frequency of 3–8 Hz. Fluctuation of fundamental frequency is maximum at 5 Hz in low-middle pitch and 5–8 Hz in high pitch.

ED = embouchure dystonia; ET = embouchure tremor; MD = musician dystonia; FTSD = focal task-specific dystonia.

tongue dystonia and Meige phenotype.[14] Meige syndrome has also been reported as a consequence of spread of ED.[7,8,14] Fifteen percent of patients reported dystonia while performing other oral tasks. Thirty-eight percent of patients with jaw, tongue dystonia, and Meige phenotype reported dystonia while speaking.[14] However, this spread of dystonia is not observed in lip-locking and ET.[7] Coexisting FTSD of hand has also been reported[20] with few musicians developing focal hand dystonia many years (up to 19 years) prior to the onset of ED, indicating the possibility of genetic predisposition.[8]

Geste Antagoniste and Pain in ED

Geste antagoniste, reflecting the effect of internal sensory input on the sensorimotor plasticity, is rarely observed in ED.[20] Geste antagoniste was seen in 10% of patients with ED in one study.[14] However, when it is effective, the response is almost dramatic.[20] Cooling of periorbital area has resulted in transient improvement in ED in a tuba player.[22] Although pain is characteristic in embouchure disorders such as Satchmo syndrome, pain is observed in 12%–16% of patients and oral discomfort is observed in 35%–42% of patients with ED.[8,14]

Hand Dystonia in ED

Wind instrument players have been reported to develop musicians' hand dystonia.[19,23] Flutists engage in more complex finger movements hence explaining higher chances of focal hand dystonia compared to the brass musicians.[24] In one case report, a Nadaswaram (an Indian double-reed instrument made of wood

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Table 2: Phenomenology of focal task-specific lower limb dystonia

Authors	Patient	Findings
Lee et al., 2014[37]	Percussionist aged 28 years	Age of onset – 26 years. Tightness of the right thigh while performing rapid, repetitive, and tapping movement of foot. EMG analysis showed co-contraction of quadriceps and biceps femoris. In addition, tibialis anterior and gastrocnemius showed alternate contraction in both affected and non-affected legs.
Katz et al., 2013[35]	Percussionist aged 75 years	Age of onset – 45 years Plantarflexion of right foot.
Altemuller et al., 2012[24]	FTSD in 591 musicians compared to 2,651 healthy musician students	FTSD of lower limb in four percussionists. Percussionists were more likely to have lower limb FTSD compared to other musicians.
Rosset-Llobet et al., 2012[36]	Percussionists aged 22 and 23 years – age of onset 20 and 23 years, respectively	Patient 1: age of onset – 20 years Sensation of tension over muscles of left foot, ankle, and knee causing blocking of knee while playing drum. Patient 2: age of onset – 23 years Impairment of alternate flexion and extension of both feet. In addition, while playing, toes underwent flexion and there was elevation of heel. In both patients, EMG showed co-contraction of anterior tibialis and gastrocnemius.
Asahi et al., 2018[70]	Percussionist aged 37 years	Age of onset – 22 years Cramping of right hand and loss of control of right lower limb while drumming.

EMG = electromyogram; FTSD = focal task-specific dystonia.

with metal at the end of the instrument) player with ED initially presented with ET and task-specific upper limb tremor but later developed postural tremor of upper limb.[25] The laterality of involvement is dependent on burden on the fingers.[24,26]

Differential Diagnosis of ED

Common complaints in ED include difficulty in playing, inability to control the movement of embouchure, tremor, and fatigue of lips.[8] ED needs to be differentiated from other disorders affecting the embouchure. Commonest non-dystonic affliction of embouchure includes infraorbital neuropathy, Satchmo syndrome (orbicularis oris tear), and overuse syndrome. Infraorbital neuropathy results in sharp, localized pain over cheek while playing an instrument and is occasionally associated with paraesthesia. Overuse syndrome is characterized by pain over cheek, fatigue, and numbness while playing. In Satchmo syndrome, the presentation is acute onset of pain, numbness, and difficulty in playing an instrument.[7] Mechanical injury to the embouchure also needs to be ruled out.[20] Stress-induced velopharyngeal incompetence characterized by air leak through nose while playing an instrument has been described.[27] Playing instruments requiring high intraoral pressure is associated with this disorder.[28,29] The symptoms develop 30 minutes after playing the instrument.[27]

Diagnosis

Evaluation of ED is difficult due to challenges in visual inspection, especially when tongue and larynx are involved, unlike FTSD of hand that is readily visible.[30] Evaluation of patients with ED is augmented when musicians are examined while buzzing (an exercise wherein musicians vibrate their lips while performing embouchure tasks), while using an O piece (a device which allows the musician to observe their own lips while practicing), and when musicians are using only the mouthpiece of the instrument.[20] Palpation of the embouchure is essential to look for tenderness or nodules in order to rule out other causes of embouchure discomfort.[7] Quantification of severity of ED can also be attempted based on acoustic measures[30,31] (Table 1). On studying the acoustic analysis, high registers are most severely affected.[30,32]

ED carries a high risk of relentless progression and high chances of spread of dystonia to other tasks particularly in jaw phenotype.[7,8,14] Hence, musicians with ED must be strongly advised to discontinue playing the instrument.[14]

Lower Limb Dystonia

FTSD in Drummers

Epidemiology. The exact incidence of FTSD of lower limbs among drummers is not known. In a study of 591 instrumentalists with FTSD, FTSD of lower limb was found in 4 musicians. They were more likely to be percussionists according to the study.[33] Lederman reported musicians' hand dystonia in 6 percussionists out of 139 musicians with dystonia.[34] However, none of these patients had lower limb drummers' dystonia. Four drummers with FTSD of lower limb have been described in other series.[24,35,36]

Precipitating Factors

In a case series of lower limb drummers' dystonia, the onset of dystonia closely followed a recent modification of practice,[36] which is a well-known trigger factor in musicians' hand dystonia.[17,19] In another case series, the drummer developed symptoms when he began playing double bass pedal (alternate foot movements were used to push respective pedals for drum percussion).[37]

Phenomenology

FTSD of lower limb among drummers has been commonly observed in distal lower limb.[36] However, FTSD of proximal

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lower limb muscles has also been reported.[37] Symptoms reported by these individuals include impaired dexterity, incoordination, and loss of rhythmicity of foot movements while playing the pedal, difficulty in performing alternate foot movements, sensation of involuntary tension over knees, legs, and ankles, and tightness of thigh muscles. Involuntary posturing of toes and feet and locking of knees were also reported. Symptoms were aggravated when the beat frequency was high and during alternate foot movements. Symptoms relieved with lowering the rapidity of limb movements and unilateral limb movements.[37] Katz et al. reported beneficial effects of geste antagoniste in alleviating FTSD in a drummer. The drummer placed the toes on a higher surface and used the heel instead, for drumming[35] (Table 2).

Spread of dystonia to other tasks has been seldom reported in FTSD of lower limb of drummers. Only one case series narrates an account of a drummer who reported inward twisting of left toe during ambulation.[35]

Diagnosis. FTSD of lower limb needs to be differentiated from paroxysmal kinesigenic dyskinesia. Dystonia in paroxysmal exercise-related dyskinesia develops after few minutes of exertion unlike FTSD that develops immediately after initiation of task.[36] Routine electromyogram is normal in FTSD of lower limbs and is essential to rule out myogenic and neurogenic causes of lower limb movement impairment. However, electromyogram performed during task performance gives way to the diagnosis.

Electromyogram performed while performing the task shows contraction of agonist-antagonist (anterior tibial and gastrocnemius muscles) of lower limb[36,37] (Table 2). In the patient with proximal lower limb involvement, electromyogram (EMG) revealed overflow of activity into the thigh resulting in co-activation of both quadriceps and biceps femoris muscles.[37]

Singer's Dystonia. SD finds its mention particularly in otorhinolaryngology practice. SD is a rare task-specific dystonia in singers with features of spasmodic dysphonia while singing only. While speaking, voice remains normal in these individuals[38,39] This is in contrast to spasmodic dysphonia, wherein the dystonic element affects the vocal cord when the patient is speaking. The voice is normal in individuals with spasmodic dysphonia when they are singing or laughing.[40]

Phenomenology. Symptoms of SD include cracking of voice at a particular pitch, instability at a particular register, difficulty in producing notes at a particular pitch with one patient reporting voice breaks, and requiring to cough and take a breath to continue. Patients also complain of breathless quality of voice when singing at a particular pitch. Voice fatigue while speaking has been reported.[40] Other symptoms include unsteadiness, tightness of voice while singing certain ranges, loss of vibrato (slight variation of pitch while singing), and resonance.[39] One of the patients suffered from vocal tremor and torticollis.[40] Fabiani and Teive reported a 24-year-old singer who developed alteration of pitch and cervical discomfort while singing that progressed to cause left torticollis with laterocollis, implying the coexistence of SD and task-specific spasmodic torticollis.[41] Spread of the dystonia to involve speech resulting in spasmodic dysphonia has also been reported. Family history of similar complaints was reported by one patient.[39,40]

Diagnosis. SD poses a diagnostic challenge. It can be mistaken for improper singing technique or technical issues such as laryngeal and tongue tension or difficulties during register shift.[40] A vocal register indicates a specific set of tones or pitch with similar voice quality that can have varied ranges. Traditionally, three voice registers have been recognized – head voice, chest voice, and middle voice.

Laryngeal EMG in SD reveals dystonia of posterior cricoarytenoid muscles and thyroarytenoid muscles. Adductor-type task-specific dystonia is more common than abductor type of dystonia.[39,40] The diagnosis of SD requires (a) task-specific (i.e., singing a particular note) voice instability characterized by alteration in tone and vibrato, which may or may not be associated with pitch specificity, (b) spectrographic analysis of singing samples that reveals aphonic breaks on performing abductor tasks, widely separated vertical striations on performing adductor tasks, and (c) laryngeal EMG that displays increased baseline activity and abnormal discharge of thyroarytenoid and posterior cricoarytenoid muscles at the affected pitch and at rest.[40]

SD shares similarities with musicians' hand dystonia in terms of task specificity, affliction of small group of muscles, propensity to spread to other tasks, and its existence among musicians who have dedicated many years to vocal practice. Register specificity is similar to that seen in ED, seen in French horn players.[20] However, certain characteristics differentiate it from musicians' hand dystonia. Unlike musicians' hand dystonia or lower limb dystonia, SD does not appear to trigger with a change in singing patterns or recent increase in voice exertion. However, these individuals had spent many hours in practicing singing. This itself could be a predisposing factor for SD.[40]

Pathogenesis. In a seminal article on the pathogenesis of task-specific dystonia, Byl and co-workers reported that repetitive stereotyped movements might facilitate maladaptive reorganization in the brain that might result in dystonia instead of perfection of the movements.[42] They substantiated this statement by performing cortical mapping of adult monkeys exposed to repetitive hand stress. They found an increased overlap in the receptive fields of the skin indicative of degradation of normal precise representations of various areas of the hand.[42] Similar results of loss of differentiation and reorganization of somatosensory maps were replicated in a study of musicians' hand dystonia.[43,44]

Many theories have been proposed as the pathogenetic mechanisms behind musicians' hand dystonia. These include abnormal plasticity[45] leading to enlarged cortical representation[46] and enhanced undifferentiated excitability of the entire hand motor cortex.[47] The aberrant plasticity of the cortical-striato-pallido-thalamo-cortical circuit in dystonia[48] might lead to overactivation of the premotor cortex that is seen in musicians' hand dystonia.[49] Weak intracortical inhibition[50,51] and low gamma amino butyric amino acids levels in motor cortex[52] explain the mechanism of abnormal cortical excitability.

Pathophysiological Differences in Task-Specific Dystonia of Musicians and Nonmusicians. Comparative studies of FTSD among both musicians and nonmusicians emphasize the differential involvement of motor programs that results in dystonia. A study presented the concept of abnormal organization of network kernel that was representative of altered connectivity in FTSD.

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Table 3: Pathophysiological insights in ED through imaging

Technique	Authors	Study population	Findings
Functional magnetic resonance imaging	Uehara et al., 2018[61]	Wind instrumentalists with ED (n = 14) and healthy wind instrumentalists (n = 14)	In right primary somatosensory region, significantly decreased intracortical distance between hand and mouth cortices was observed. There was no difference in sensorimotor cortex activity between ED and healthy controls. Primary somatosensory, primary motor, putamen, and cerebellar activation were coherent with greater fundamental frequency variability seen in ED.
	Mantel et al., 2016[59]	Brass players with ED (n = 15) and brass players without ED (n = 15)	Overactivation of cerebellar sensorimotor area following stimulation of ipsilateral dystonic upper lip was seen in patients with ED. Primary somatosensory area showed ipsilateral overactivity on stimulation of dystonic upper lip and contralateral overactivity on stimulation of non-dystonic forehead. Bilateral secondary somatosensory overactivity was seen mainly over opercular region.
	Haslinger et al., 2017[60]	Brass players with ED (n = 30), matched with healthy musicians	Functional connectivity was altered over bilateral sensorimotor region (mouth and hand cortex), right, mesial frontal region, second somatosensory region, cerebellum, and auditory functional network.
	Haslinger et al., 2010[58]	Brass players with ED (n = 10) and brass players without ED (n = 10)	Bilateral premotor, primary motor, primary and secondary somatosensory cortex (areas of mouth representation), and mainly right primary cortex showed increased activation both during ED and at rest, more during performance of oral tasks. There was no correlation between the severity of task-induced dystonia and degree of activation of sensorimotor cortex inferring that overactive sensorimotor cortex is an intrinsic abnormality acting as a predisposing factor for the development of task-specific dystonia.
	Frucht et al., 2009[14]	Musicians with ED (n = 89)	No differences in sensory perception were found among patients with ED, healthy musicians, and nonmusicians. Lower lip and index finger show relatively more defined sensory perception in comparison with upper lip.
	Hellwig et al., 2019[55]	Real-time MRI of oral cavity in trumpet players with ED (n = 4) and healthy trumpet players (n = 10)	Increased variability of anterior tongue in ED
	Iltis et al., 2019[57]	Real-time MRI of oral cavity of healthy French horn players (n = 6), French horn players with ED	Tongue elevation was lower in musicians with ED. They also had decreased size of oral cavity while transitioning notes.
	Iltis et al., 2015[56]	Real-time MRI of oral cavity of brass musicians (n = 4)	Forward movement of tongue was observed resulting in decrease in the size of posterior compartment in trombone and tuba players. There was also increase in size of oropharyngeal area in trumpet players.
Voxel-based morphometry	Mantel et al., 2019[65]	Brass musicians with ED (n = 24), healthy brass musicians (n = 23), and healthy nonmusicians (n = 24)	Gray matter volume was increased over pre- and post-central gyrus as well as sulcus over the right side and left precentral gyrus in ED. Basal ganglia and thalamic symmetry are increased in ED compared to nonmusicians.

ED = embouchure dystonia.

They studied differences between SD and spasmodic dysphonia as well as writer's cramp and musicians' hand dystonia. They concluded that an important fact distinguishing FTSD among musicians and nonmusicians is the recruitment of subcortical region, that is, cerebellum and thalamus in FTSD of musicians.[53] Bianci et al. reported that reduced functional connectivity in parietal operculum, primary somatosensory cortex, supplementary motor area, and superior and inferior parietal lobule is an underlying trait that differentiates the task-specific nature of dystonia in musicians (SD and musicians' hand dystonia) from non-MD (spasmodic dysphonia and writer's cramp).[54]

Embouchure Dystonia. The instrument-specific nature of ED indicates that maladaptive plasticity is inflicted upon a specific motor program.[8] An example would be the normal motor program of lip closure that develops aberration culminating into an abnormal motor program of lip-locking dystonia.[8] While playing high-register instruments, high intraoral pressure builds up. To compensate for this pressure, embouchure abnormalities may develop.[8] Abnormalities in the oral cavity and tongue dynamics while playing embouchure are observed in ED[55–57] (Table 3). These faulty techniques could be related to the faulty motor programs that develop in ED. Further studies are required to

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Table 4: Pathophysiological insights in ED through electrophysiological studies

Author	Aim of the study	Subject population	Findings
Bulica et al., 2019[67]	To delineate functional connectivity in a patient with ED using MEG-CSI	Professional female flutist (n = 1)	Coherence was increased in bilateral parietal and inferior frontal region indicating that the sensory homunculus is distorted in ED.
Hirata et al., 2004[66]	To study the somatosensory representation of lips in ED using MEG	Professional male brass musicians with ED (n = 8)	Musicians had lower lip sensitivity compared to that in fingers. The sensitivity was decreased in upper lip compared to lower lip. The cortical digit representation was shifted towards the lip representation in ED.

MEG-CSI = magnetoencephalography-coherence source imaging; ED = embouchure dystonia.

decipher whether this is a consequence of ED or it is the faulty practice technique that makes a musician susceptible to aberrant motor program and subsequently to ED.

Several hypotheses have been proposed to explain the pathogenesis of ED. One of the suggested theories is aberrant plasticity of sensorimotor cortex. Functional magnetic resonance imaging (MRI) studies in ED have shown overactivity in sensorimotor region and premotor region,[58] as well as parietal operculum.[59] Similar results have been replicated in resting-state MRI studies with demonstration of altered functional connectivity in bilateral sensorimotor region over clinically asymptomatic hand cortex, in addition to oral region.[60] The mechanism explaining this finding was maladaptive organization of sensorimotor cortex and altered cortical hyperexcitability.[60] The inappropriate sensory gating by an overactive basal ganglia resulting in abnormal interaction of the somatosensory and motor input is another proposed hypothesis.[58]

Structural and connectivity abnormalities of cerebellum have been implicated in ED,[61] akin to focal hand dystonia.[62–64] In ED, in addition to involvement of primary motor cortex, primary somatosensory cortex, and putamen, activity within cerebellum was associated with dystonic features.[61] Gray matter changes[65] and distorted homunculus[66,67] have also been described in ED (Tables 3 and 4).

Upper Limb and Lower Limb Involvement in FTSD. Many factors explain the predisposition of the hand for FTSD. Studies on somatotropic organization of striatum revealed a greater degree of overlap in the hand and mouth areas compared to that of foot. Another possible explanation could be that the area for motor control of hands and face is larger than that of leg.[68]

The rarity of lower limb task-specific dystonia in comparison with the upper limb dystonia could also be due to differential degree of plasticity that occurs in the motor cortex.[69]

Bassists have not been reported to have musicians' hand dystonia. They play widely separated notes and hence simultaneous activation of digits while playing seldom occurs. This could prevent overlapping and blurring of cortical representation of digits that are implicated in musicians' hand dystonia.[19] A similar concept could explain the low incidence of lower limb dystonia. Percussionists have comparatively more focused muscle group activity involving the ankle and hence have a higher predisposition toward dystonia of lower limb. When the focused task becomes concentrated to a smaller group of muscles and becomes intense, the artists are at risk of developing FTSD.

Brain Changes in SD. In a study that compared the functional and structural aberrations among various FTSDs, compared to focal hand dystonia, patients with SD had gray matter volume abnormalities over the cortical representation for the larynx, that is, left superior parietal lobule, right inferior frontal gyrus, and anterior insula. In addition, white matter integrity was impaired in the ventral area of the precentral gyrus in SD and hand area of precentral gyrus in focal hand dystonia.[54] Another study also found that superior parietal cortex is involved in laryngeal dystonia. They also reported prominent network connectivity between the parietal cortex and frontal and occipital regions in patients with laryngeal dystonia. Superior parietal cortex is essential for higher-order sensorimotor integration. Hence, in patients with laryngeal dystonia, cortices involved in sensorimotor processing are preferentially affected, while focal hand dystonia is characterized by abnormal representation of motor execution cortices that include primary motor cortex and cerebellum.[53]

Conclusion

ED, FTSD of lower limb in drummers, and SD, although rare, cause significant morbidity among performing artists. Misdiagnosis has been frequently reported in the literature, more so in ED and lower limb dystonia. The symptoms have been frequently ascribed to have musculoskeletal or psychogenic basis. Careful inspection aided with surface EMG will help in diagnosis. Further functional studies will be able to ascertain the differential involvement of motor programs in FTSD of lower limbs in percussionists and nonmusicians.

Conflicts of Interest

There are no conflicts of interest.

Statement of Authorship

Acquisition and interpretation of data and drafting of the review were undertaken by SR. Concept and design of the review and revisions of the manuscript were undertaken by PKP.

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