ORIGINAL ARTICLE

Task-Specific Dystonia in Professional Musicians

A Systematic Review of the Importance of Intensive Playing as a Risk Factor

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SUMMARY

Background: Focal dystonia in professional musicians is a movement disorder that manifests itself during playing. It is a multifactorial condition in which a genetic predisposition and exogenous factors both play a role. Evidence suggests that intensive playing is a risk factor for the development of task-specific dystonia in professional musicians.

Methods: This review is based on pertinent publications (1950–2013) retrieved by a systematic search in medical and musicological databases. The references of the retrieved publications were also considered in the search.

Results: 16 articles with clinical information on a total of 1144 affected musicians were reviewed systematically. Their overall quality was intermediate to poor, and a meta-analysis was therefore not possible. The Bradford Hill criteria were applied to study a possible causative link between intensive playing and musician's dystonia. Musician's dystonia generally arises after at least ten years of intensive playing (corresponding to roughly 10 000 hours of practice). An association was found between the affected limb and the type of instrument: the limb that is subject to the greatest fine motor demands is the one most commonly affected. The average age of onset is 28 to 44 years.

Conclusion: The Bradford Hill causality criteria indicate that intensive playing is related to the development of musician's dystonia. In particular, the association of the type of instrument with the site of dystonia supports this thesis. The findings imply that task-specific dystonia in professional musicians should be included in the list of occupational diseases in Germany.

► Cite this as:

Rozanski VE, Rehfuess E, Bötzel K, Nowak D: Task-specific dystonia in professional musicians—a systematic review of the importance of intensive playing as a risk factor. *Dtsch Arztebl Int* 2015; 112: 871–7.
DOI: 10.3238/arztebl.2015.0871

Dystonia in professional musicians is a task-specific focal movement disorder that is accompanied by a loss in fine motor control while playing a musical instrument (1, 2). Cramping, flexions, and hyperextensions of the affected limb have been observed (2). About 1% of all professional musicians are afflicted (2). For 25% of this group, the occurrence of the symptoms means the end of their professional careers (3).

Therapeutic methods include injecting botulinum toxin, anticholinergic medication, and modifying the instrumental technique in order to change sensorimotor processing (4). Recommended measures in order to prevent musician's dystonia include proprioception training, regular breaks, and stress management (5).

Disorders that are related to musician's dystonia are further task-specific focal movement disorders, the most common of which is writer's cramp (6); other pathologies include golfer's cramp (7), auctioneer's cramp (8), or cramp while playing table tennis (9), but these are rarer.

Pathophysiologically, dystonia in musicians has a multifactorial genesis, whereby a genetic predisposition (10) interacts with exogenous factors (2). In other forms of focal movement disorders—for example blepharospasm, cervical dystonia, or orofacial dystonia—an increase in mechanical strain on the affected regions before manifestation of dystonia symptoms has been described (11). This observation has given rise to the hypothesis that intensive playing is a pathogenetically relevant risk factor for the development of dystonia in musicians, as has already been expressed in some studies (1, 12).

This systematic review aims to investigate the importance of intensive playing as a risk factor for the development of focal, task-specific dystonia in professional musicians. To this end,

- we compared the prevalence between an exposed group of persons and the general population without exposure, and
- we studied a possible association between instrument-specific demands on fine-motor precision and affected body parts.

Methods

Search strategy

Initially we established a review protocol, which can be accessed online (13). We conducted a systematic

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literature search in 23 medical and musicology databases (*eTable 1*). Furthermore, other data sources, such as Google and Google Scholar were searched for relevant publications for the time period from January 1950 to April 2013. Online publications were included, as were abstracts and presentations from conferences. We used the following search terms: ["musician" OR "performing artist" OR "pianist" OR "cellist" OR "drummer" OR "woodwind" OR "flutist" OR "violinist"] AND ["dystonia" OR "focal dystonia" OR "cramp" OR "embouchure" OR "musician's dystonia" OR "occupational dystonia"]. In databases allowing users to only enter one search term, an abridged search strategy was employed. In German-language databases, the equivalent German terms were used. Additionally, we searched the bibliographies of the included research studies and review articles for suitable publications.

In line with the objective of this systematic review, we defined the following tasks:

- To undertake a comparison of prevalence rates of focal (task-specific) dystonia between a group with exposure and the general population
- To conduct an instrument-specific analysis of the anatomical manifestation of focal dystonia in affected musicians.

We assumed that the body part that carries the main burden of fine-motor strain and in which gross motor strength and fine-motor coordination interact will be most commonly affected by dystonia. If this hypothesis is true, different body parts within the different classes of instruments are likely to be affected.

The PICOS criteria (14) were defined as follows:

- P (population): professional musicians
- I (intervention): intensive playing
- C (comparison group): the non-professional normal population who makes music
- O (outcome): a) anatomical manifestation of focal, task-specific dystonia within predefined instrument groups; b) conclusions regarding the prevalence of dystonia in musicians
- S (study design): systematic review of original articles that include clinically documented cases.

Inclusion criteria

Articles were regarded as suitable for inclusion—independently of the study design—if they contained clinical and epidemiological primary data on musicians with focal task-specific dystonia. The diagnosis would have had to be made by a neurologist, and the article had to include information on the affected body part, the instrument/s played, and the intensity of instrument practice.

After deduplication, one of the authors (VR) checked the title, abstract, and full-text article for their suitability, and non-relevant articles were excluded. We chose primarily German and English as the publication languages. For other languages, articles were checked by interpreters in order to find out whether they contained clinical information in affected musicians.

Exclusion criteria

Our exclusion criteria were: doubts in the confirmation of the diagnosis by an expert and lacking clinical information.

Data extraction and quality assessment

Information from the included studies was collated by using a pre-prepared data extraction form. The following data were collected:

- Publication date (author, year of publication)
- Study design
- Setting (country, city, hospital)
- Primary endpoint (number of affected patients)
- Age of subjects at the time of illness onset
- Sex
- Affected body part
- Exposure (instrument/s played, quantification of playing)
- Factors accompanying the disorder or prior factors (especially psychological stresses, prior trauma, hereditary predisposition).

The instruments were grouped into six classes; the classes reflected the anatomical distribution of the strongest fine-motor strain/stress (1):

- Keyboard instruments (piano, organ, accordion),
- String instruments (violin, viola, cello),
- Plucked string instruments (harp, banjo, guitar),
- Woodwind instruments (flute, clarinet, saxophone, oboe),
- Brass instruments (trumpet, trombone, horn)
- Percussion.

Where necessary, a study's authors were contacted for additional information.

The quality of the included studies was assessed by using the Liverpool modification of the Newcastle-Ottawa-Scale (15). This approach was chosen for its flexibility, because it makes it possible to evaluate different types of studies and to adapt the response categories to the research question of this systematic review. Furthermore, this assessment scale allowed us to assess the cohort under study, the exposure, a study's primary result, and the studies' results. Up to two points per category are allocated; in total, 16 points can be awarded (*eTable 2*).

Data extraction and quality assessment were carried out independently by two authors of the present study (VR and either ER, DN, or KB) (*Figure 1*).

Data evaluation

The prevalence rates of manifest focal dystonia depending on the main instrument played—grouped by the respective class of instrument (keyboard instruments; plucked string, string, woodwind, brass, and percussion instruments)—were graphically combined for all included patients (*Figure 2*).

Since some of the studies with smaller case numbers focused on very specific research questions and because this might lead to selection bias, a sensitivity analysis was undertaken, and only those studies were considered that included more than 100 patients. In

order to be able to categorize the relationship between intensive playing and dystonia symptoms as causal or merely associative, we used the Bradford Hill criteria for causation as the basis.

Results

In total, we identified 866 articles, of which 16 (1, 16–30) met the inclusion criteria (*Figure 1*). All included studies were case series or clinical-experimental studies, with case numbers between three and 591 (*eTable 3*). Four studies were large case series (>100 affected musicians) that had been conducted at specialized centers for musicians' medicine in Germany, France, Spain, and the US (1, 16, 28, 31). These four studies included data on 930 patients in total. Twelve studies were small and reported on 214 cases. According to the quality assessment, the studies were of poor to moderate quality (between 6 and 10 points out of 16) (*eTable 3*).

From the included articles we were able to identify a mean age at manifestation of 28–44 years. More men were affected than women (57–100%). The intensity of practicing was reported in different ways: as a daily practice time of four to six hours (22, 27, 28), as a cumulative total practice time of about 10 000 hours or more (20), or as a practice period of 10 or more years (26). Detailed information on the familial predisposition of musician's dystonia was not available. Reports on the frequency of dystonia in the included articles were based either on literature citations or on primary

FIGURE 1

Selection process and inclusion of suitable studies into the systematic review according to the PRISMA criteria

data from a center for musicians' medicine (*Table 1*), and these can therefore not be used as frequency data. Further inquiries yielded the insight that the prevalence data for Germany consist of the estimated number of affected musicians at specialist centers (ca 800) and the total number of professional musicians registered in orchestral associations (about 80 000). This equates to a

TABLE 1

Prevalence rates of dystonia in musicians as reported in included studies

Author, year (literature)	Country	Prevalence data	Data collection
Altenmüller et al., 2010 (review) (2)	Germany	1% of all professional musicians	Cited (Altenmüller) (e1)
Aranguiz et al., 2011 (review) (e2)	Spain	0.5–1% of all professional musicians	Cited (Jabusch [39], Pullman [e11])
Conti et al., 2008 (review) (17)	USA (New York)	In up to 1 in 200 cases; 8–14% in specialist hospitals/clinics	Cited (Hochberg, Lederman) [24, e3])
Frucht et al., 2001 (19)	USA (New York)	Embouchure dystonia: up to 8% specialist clinics	Cited (Knishkowsky) (e4)
Jankovic et al., 2008 (review) (e5)	USA (Houston)	8% of all musicians	Cited (Lederman) (e12)
Lederman, 2003 (e6)	USA (Cleveland)	8% of all musicians from authors' own specialist clinic	Primary data
Rosset-Llobet et al., 2009 (e7)	Spain	13% of all musicians (87 out of 665 musicians who attended a specialist clinic)	Primary data
Lie-Nemeth, 2006 (review) (e8)	USA (Boston)	5% (cited Brandfonbrener) (e9) 8% (cited Lederman) (e6) 11% (cited Tubiana) (31) 13% (Newmark, Hochberg) (e10)	Citations relate to data from specialist clinics (personal communication from musicians' outpatient clinic)
Brandfonbrener, 1995 (e9)	USA (Chicago)	58 patients with musician's dystonia out of a total of 3918 patients seen = 1.5%	Primary data from a specialist clinic
Tubiana, 2003 (31)	France (Paris)	145 patients with focal dystonia out of a total of 4000 musicians = 3.6%	Primary data from a specialist clinic
Altenmüller et al., 2012 (1)	Germany (Hanover)	591 musicians with dystonia out of 5500 patients attending a specialist hospital/clinic/department (author's personal communication) = 10.7%	Primary data from a specialist clinic

Distribution of focal dystonia by instrument group in the 1144 musicians included in this review

FIGURE 2

cumulative prevalence of 1% (Altenmüller, personal communication) (1, 5).

For the reported prevalences of idiopathic focal dystonia we used a large prevalence study (32) and a systematic review (33). These studies found rates of 117 per 1 000 000 (1 : 8500) (32) and 15 per 100 000 (1 : 6600). Estimated prevalences for limb dystonia in the group of non-professional musicians in the normal population are 1.24 per 100 000 (33).

Among musicians playing keyboard instruments ($n = 327$), the dystonia manifested preferentially in the right hand (dystonic symptoms in the right hand: 78%; left hand: 15%; both hands: 6%; torticollis: 1%). This was also the case for musicians playing plucked string instruments ($n = 245$; right hand: 78%; left hand: 19%; both hands: 3%) (Figure 2). The left hand was mainly afflicted in musicians playing string instruments ($n = 155$; right hand: 68%; right hand: 30%; both hands: 1%; torticollis: 1%). Orofacial dystonia—also called embouchure dystonia—was found exclusively in players of woodwind and brass instruments (woodwind: $n = 199$; 26%; brass: $n = 181$; 96%). Percussionists showed an almost equal distribution between both hands (left hand: 41%; right hand: 49%; both hands: 2%), but—in contrast to all other musicians—they also developed symptoms in their feet (8%).

These results were confirmed in a sensitivity analysis exclusively of the data of the four large studies (1, 16, 28, 30) (eFigure).

Discussion

Focal, task-specific dystonia in professional musicians is a multifactorial disorder in which—according to the

current hypothesis regarding pathophysiology—exogenous factors add to a genetic predisposition (1, 2). In this systematic review we investigated the importance of intensive playing as a trigger factor for the disorder. On the one hand, we found a notably increased prevalence of the disorder among the group that had been exposed compared with the non-exposed normal population; on the other hand, however, we also found instrument-specific manifestations of dystonia symptoms.

A comparison of the prevalences of focal idiopathic dystonia in the normal population with dystonia in musicians shows clear differences of 1:6600 to 1:100. With regard to focal, task-specific movement disorders, we even found prevalence differences of 1:80 000 to 1:100. Furthermore, it is of note that task-specific dystonia during instrument practice has been reported in the literature only for professional musicians, but not for the normal population—that is, lay musicians.

The disorder that is most closely related to musician's dystonia is writer's cramp, a similarly task-specific movement disorder. A case-control study identified as the risk factors long years of extensive writing as well as an abrupt increase in the duration and intensity of writing (6). An increase in the intensity of instrument practice before the manifestation of dystonia symptoms has also been observed in affected musicians (17), but this has not been the subject of systematic study. A case-control study was conducted for musician's dystonia (34); this identified an older childhood age when taking up playing an instrument as a risk factor.

When analyzing the anatomical distribution of dystonia symptoms by specific type of instrument, it is of

TABLE 2

Bradford Hill criteria and their applicability

Bradford Hill criterion	Applied in the setting of dystonia in musicians
Strength of the association	Prevalence comparison between exposed and non-exposed groups: 1 : 100 versus 1 : 6600 / 1 : 8500
Consistency	Comparability of results in different cohorts, here: similar results reported in studies from different countries
Specificity	Direct association between exposure and outcome is obvious, here: the manifestation of dystonia symptoms specific to the instrument class; exclusive occurrence of foot dystonia in percussionists and orofacial dystonia in brass instrumentalists.
Temporality	The exposure has to precede onset of the disorder; here: intensive music-playing precedes the occurrence of dystonia in musicians.
Biological gradient	Relation between exposure and manifestation; a minimum playing time of 10 practice years/10 000 practice hours has been described as a possible threshold.
Plausibility	The association between exposure and manifestation of dystonia seems logical; here: the instrument-specific manifestation of the dystonia symptoms is consistent with the main fine motor stress within the respective instrument class.
Coherence	Substantial agreement between epidemiological facts and laboratory-based experimental discoveries; here: induction of focal hand dystonia by repetitive hand movements in monkeys (e13, e14)
Experiment	Reduced exposure leads to alleviation of symptoms: improvement of dystonia as a result of playing very slowly (36)
Analogy	Similarities to other forms of exposure; here: analogy to writer's cramp, which is also associated with local exposure.

note that the body part that is subjected to the highest degree of fine motor stress/strain is also the body part that is proportionally most often affected. The dominance of the right hand in musicians playing keyboard instruments or plucked string instruments and the left hand in musicians playing string instruments that this study identified is consistent with the mechanical challenges within these classes of instruments (1, 12). Brass instrumentalists and musicians playing plucked string instruments were at a particularly high risk for developing dystonia (1); a fact that may reflect the enormous mechanical challenges posed by these types of instruments.

This systematic review underlines the importance of intensive playing as a trigger factor. The decision of whether two variables are linked by causation or by association can be based in the epidemiological setting on the Bradford Hill criteria (35). *Table 2* provides a complete listing of all nine criteria.

Both research questions addressed in the present study were directed at the criteria “strength of the association” and “specificity.” The strength of an association means that a causal association is more likely when the risk of developing a disorder in the exposed group is substantially higher than in the non-exposed group. The specificity expresses that the selective effect of the exposure—in this case, more intense music practice—increases the risk of the disorder; this is also supported by the fact that the manifestations of dystonia are instrument-specific. The other criteria are also well explained in the case of musician's dystonia (*Table 2*), so that the assumption of a causal association between intensive playing and the manifestation of dystonia symptoms is justified.

Detailed knowledge regarding the type and duration of the exposure are important for the possible formal recognition of focal dystonia as an occupational disorder, as well as for the development of prevention and rehabilitation measures.

Quantifying exposures is difficult in view of the available data. In terms of the medical history, a minimum playing time of 10 practice years (26) has been reported; this corresponds to cumulatively 10 000 hours of musical practice (20) or six hours a day (22). Furthermore, an increase in the intensity of practice has been reported within the year preceding the initial manifestation of dystonia symptoms (17).

Different approaches have been reported for the purpose of modifying exposures. These include:

- Playing very slowly (36)
- Wearing a splint (37)
- Learning Braille script for blind people in order to change sensorimotor processing (38).

Some 50% of affected musicians benefited from re-training strategies (4). Rates of occupational invalidity/disability varied between 25% (3) and 50% (39).

Limitations

This systematic review and its results are subject to unavoidable weaknesses and limitations that have to be considered for the purposes of their interpretation. The literature search was not conducted independently by two reviewers. The quality assessment of the included articles identified studies of poor to moderate quality, which was mostly due to the fact that the research was clinical, not (prospective) epidemiological research, with the relevant study design and consideration of

biases. Especially exposure data were based on patients' medical histories and retrospectively collected data. We were, however, able to identify a large number of cases with good quality clinical documentation of their disorder. Studies from different countries showed a consistent overall picture regarding focal dystonia in professional musicians.

Conclusion

This systematic review shows a clear and potentially causal association between intense musical practice and the manifestation of focal dystonia in professional musicians. This means that, in our estimation, the scientific requirements for including focal, task-specific dystonia among professional musicians in the catalogue of occupational disorders are essentially provided.

We furthermore make a plea to generate the relevant health awareness among musicians, whereby they play their instruments ergonomically and take breaks, as well as avoid stereotypical and repetitive exercises.

Clear diagnostic guidelines are desirable, since an early and appropriate diagnosis by a specialist would limit incorrect diagnoses and subsequent ineffective treatments that might entail adverse effects. Further research projects to elucidate the causes and prevention of dystonia in musicians are urgently needed.

Addendum

The medical expert advisory panel for occupational diseases in the German Federal Ministry of Labour and Social Affairs decided on 1 December 2015 to recommend to the regulator to include a new occupational disease: "Focal dystonia as a neurological disorder in musicians as a result of fine-motor activity at a high level of intensity."

Conflict of interest statement

The authors declare that no conflict of interest exists.

Funding: The systematic review that forms the basis of this publication was funded by the German Statutory Accident Insurance, DGUV research project FB-0202, www.dguv.de/Projektdatenbank/0202/4177377v1.pdf

Manuscript received on 25 April 2015, revised version accepted on 13 August 2015.

Translated from the original German by Birte Twisselmann, PhD.

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KEY MESSAGES

- The estimated prevalence of focal dystonia in professional musicians is in the region of 1:100. The prevalence of idiopathic focal dystonia in the normal population is 1:6600 to 1:8500.
- An association exists between the body part (limb, muscles of the mouth) in musicians that is most commonly affected by dystonia and the instrument played. The body part that is subject to the highest degree of fine motor stress/strain is also most often affected by dystonia.
- Orofacial dystonia occurs exclusively in players of brass instruments and is therefore to be regarded as specific to these musicians.
- According to the Bradford Hill criteria for causation, the association between intensive playing and the manifestation of musician's dystonia can be classed as causal.
- Focal dystonia in musicians can be considered an occupational disorder caused by the job; therefore, in our opinion, it meets the requirements for inclusion into the list of occupational disorders in Germany.

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Supplementary material

For eReferences please refer to:
www.aerzteblatt-international.de/ref5115

eFigure, eTables:
www.aerzteblatt-international.de/15m0871

Supplementary material to:

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Dtsch Arztebl Int 2015; 112: 871–7. DOI: 10.3238/arztebl.2015.0871

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Distribution of focal dystonia by instrument group in analogy to *Figure 2*, after performing a sensitivity analysis, which includes 930 musicians from 4 large case series

eTABLE 1

Overview of databases searched and the articles obtained through the search

Database	Number of hits	Duplicates	Unsuitable articles (title)	Suitable studies (title)	Case reports	Reviews
1) PubMed	67	2	19	31	8	7
2) EMBASE	154	26	57	51	17	3
3) MEDLINE	59	20	19	10	7	3
4) PsycINFO	53	22	28	3	0	0
5) BioMedCENTRAL	2	0	2	0	0	0
6) Cochrane	3	1	2	0	0	0
7) Scientific library of the German Federal Institute for Occupational Safety and Health	0	–	–	–	–	–
8) German Statutory Accident Insurance (DGUV) rules and regulations	0	–	–	–	–	–
9) BIOSIS Previews	2	1	1	0	0	0
10) WHO Library	5	0	5	0	0	0
11) BMJ	5	3	1	1	0	0
12) CAIRS for Music	0	–	–	–	–	–
13) British Library Sound Archive	0	–	–	–	–	–
14) Music Online	0	–	–	–	–	–
15) Wiley Online Library	34	25	6	3	0	0
16) Med Problems Performing Artists	32	24	4	4	0	0
17) Performing Arts Medicine Trust	0	–	–	–	–	–
18) Cambridge Companion Online	0	–	–	–	–	–
19) German Association for Music Physiology and Musicians' Medicine (DGfMM)	0	–	–	–	–	–
20) Performing Arts Medicine Association	0	–	–	–	–	–
21) BioMedLit	76	25	25	16	9	1
22) BioMedSearch	186	90	79	13	3	1
23) ISI Web of Science	188	123	52	13	0	0
TOTAL	866	362	300	145	44	15

eTABLE 2
Clinical epidemiological studies

Phase A: Selection of studies			
Selection bias		Response bias	
Are there indications of selection bias?		Are there indications of response bias?	
Yes	0	Yes	0
Possibly	1	Possibly	1
No	2	No	2
If "Yes/possibly", please provide reason:		If "Yes/possibly", please provide reason:	
Phase B: Determining exposures			
Instrument played		Quantifying exposures	
Gradual documentation of exposure?		Quantification of music-making?	
No documentation	0	No quantification of playing	0
General documentation	1	Quantification of playing	1
Detailed documentation of primary and secondary instruments	2	Quantification of current and previous playing times in order to estimate cumulative playing times	2
Comments:		Comments:	
Documentation bias			
Are there indications of bias while documenting exposures:			
Yes	0		
Possibly	1		
No	2		
If "Yes/possibly", please provide reason:			
Phase C: Documentation of disorder			
Assessment of dystonia		Assessment bias	
Outcome assessment by		Are there indications of bias when assessing the disorder?	
Patient themselves	0	Yes	0
Doctor	1	Possibly	1
Doctor specializing in movement disorders	2	No	2
Comments:		If "Yes/possibly", please provide reason:	

Phase D: Analysis of results

Consideration of confounders

Were confounders considered?

No	Limited or lacking consideration 0
Yes, appropriately	Crucial factors considered 1
Yes, well	Most known factors considered 2

Comments:

Phase E: Quality assessment

Overall assessment documented while considering individual aspects

Total number of points x/16

Comments:

Liverpool adaptation of the Newcastle-Ottawa Scale

eTABLE 3

Overview of all included studies, data extraction, and quality assessment

Author and country	Study type	Total number of musicians affected	Number of musicians affected by instrument group and distribution of dystonia	Age at initial manifestation (mean)	Sex	Intensity of practice	Quality assessment (maximum:16)	Investigated study variable
Altenmüller et al. (2) Germany	Large case series (n >100)	n = 591	K (n = 179): 76% RH, 14% LH, 10% BH, 1% T	4 th decade of life	M: 78% F: 2%	n. a.	9	Manifestation of focal dystonia depending on instrument class
			S (n = 81): 62% LH, 45% RH, 2% T, 1% BH					
			PS (n = 105): 76% RH, 20% LH, 4% BH					
			W (n = 113): 37% LH, 35% RH, 23% ED, 4% BH, 1% T					
			B (n = 99): 98% ED, 1% T, 1% LH					
Brandfonbrener et al. (16) USA	Large case series (n > 100)	n = 113	P (n = 15): 53% RH, 40% LH, 6% BH, 1% RF	n. a.	M: 74% W: 26%	n. a.	9	Clinical end epidemiological data from affected musicians
			K (n = 34): 74% RH					
			S (n = 15): 60% LH					
			PS (n = 20): 95% RH					
			W (n = 18): 50% LH, 28% RH					
			B (n = 13): 100% ED					
			P (n = 5): 60% RH					

Author and country	Study type	Total number of musicians affected	Number of musicians affected by instrument group and distribution of dystonia	Age at initial manifestation (mean)	Sex	Intensity of practice	Quality assessment (maximum:16)	Investigated study variable
Contri et al. (17) Italy	Clinical experimental study	n = 61	K (n = 19): 52% RH, 26% BH, 22% LH	35 years	M: 82% W: 18%	n. a.	6	Clinical end epidemiological data from affected musicians depending on instrument played
			PS (n = 20): 75% RH, 26% BH, 22% LH					
			S (n = 5): 80% LH, 20% RH					
			B (n = 3): 66% BH, 33% RH					
			W (n = 9): 56% LH, 22% RH, 22% BH					
			P (n = 7): 60% LH, 40% RH					
Ferrarin et al. (18) Italy	Clinical experimental study	n = 18	K (n = 3): 100% RH	35 years	M: 83% W: 17%	n. a.	8	Comparison between clinical analysis and objective measuring method in detecting dystonia in musicians
			S (n = 5): 80% LH, 20% RH					
			PS (n = 6): 83% LH, 20% RH					
			P (n = 2): 100% RH					
			W (n = 1): 100% LH					
Frucht et al. (19) USA	Clinical experimental study	n = 26	W (n = 14): 100% ED	37 years	M: 77% W: 23%	Age when taking up instrument: 12 years Dystonia onset 25 years later	9	Clinical examination of musicians with orofacial dystonia
			B (n = 12): 100% ED					
Granert et al. (20) Germany	Clinical experimental study	n = 11	K (n = 11): 100% RH	34 years	M: 82% W: 18%	811 h/year cumulative playing time >10 000 h	10	Imaging study (MRI) in affected and healthy pianists of voxel-based morphology of gray matter
Hayes et al. (21) Australia	Clinical experimental study	n = 3	K (n = 1): 100% RH PS (n = 2): 100% RH	33 years	n. a.	n. a.	9	Assessing the therapeutic effects of botulinum toxin on orofacial dystonia

Author and country	Study type	Total number of musicians affected	Number of musicians affected by instrument group and distribution of dystonia	Age at initial manifestation (mean)	Sex	Intensity of practice	Quality assessment (maximum:16)	Investigated study variable
Hirata et al. (22) Germany	Clinical experimental study	n = 8	W (n = 4): 100% ED B (n = 4): 100% ED	33 years	M: 100%	At least 6 h/day	9	Imaging study (MRI) of voxel-based morphometry of the cortex in musicians with orofacial dystonia and healthy musicians
Kadota et al. (23) Japan	Clinical experimental study	n = 7	K (n = 3): 75% RH, 25% LH W (n = 3): 75% RH, 25% LH PS (n = 1): 100% RH	28 years	M: 57% W: 43%	n. a.	9	Imaging study (fMRI) of activating pattern in affected musicians during repetitive movements compared with healthy controls
Lederman (24) USA	Clinical experimental study	n = 43	B (n = 43): 100% ED	35 years	M: 72% W: 28%	n. a.	8	Clinical investigation and data collection of affected musicians with orofacial dystonia
Nowak et al. (25) Germany	Clinical experimental study	n = 5	K (n = 3): 100% RH PS (n = 2): 50% RH, 50% LH	38 years	M: 80% W: 20%	n. a.	9	Analysis of movement patterns and movement strength in musicians with hand dystonia
Pujol et al. (26) Spain	Clinical experimental study	n = 5	PS (n = 5): 60% RH, 40% LH	38 years	M: 80% W: 20%	>10 practice years	9	Imaging study (fMRI) during dystonic movements
Rosenkranz et al. (27) UK	Clinical experimental study	n = 6	K (n = 3): 66% RH, 33% LH S (n = 1): 100% LH B (n = 1): 100% LH PS (n = 1): 100% RH	36 years	n. a.	2–4 h/day	8	Assessment of therapeutic attempt using sensorimotor training in affected musicians

Author and country	Study type	Total number of musicians affected	Number of musicians affected by instrument group and distribution of dystonia	Age at initial manifestation (mean)	Sex	Intensity of practice	Quality assessment (maximum:16)	Investigated study variable
Rosset-Llobet et al. (28) Spain	Large case series (n >100)	n = 101	PS (n = 47): 85% RH, 15% LH K (n = 21): 80% RH, 15% LH, 5% BH S (n = 6): 84% LH, 16% RH W (n = 11): 45% RH, 18% ED, no further information available B (n = 7): 100 % ED P (n = 2): 50% RH, 50% LH n = 7: No information available	29.3 years (M); 27.5 years (W)	M: 88% W: 12%	5 h/day	8	Examination of affected musicians for further movement disorders in addition to focal dystonia during music-playing
Schuele et al. (29) USA	Clinical experimental study	n = 21	S (n = 21): 76% LH, 24% RH	34 years	M: 86% W: 14%	n. a.	10	Long-term analysis of affected string instrumentalists
Tubiana et al. (30) France	Large case series (n >100)	n = 141	K (n = 50): 90% RH, 10% LH S (n = 21): 76% LH, 24% RH PS (n = 36): 72% RH, 28% LH P (n = 6): 44% RH, 28% LH, 28% RF, 14% LF	33 years	M: 72% W: 28%		8	Therapeutic effect on affected musicians of sensorimotor retraining program

Classes of instrument: K: keyboard instrument; PS: plucked string instrument; S: string instrument; W: woodwind instrument; B: brass instrument; P: percussion
Affected body part: RH: right hand; LH: left hand; BH: both hands; RF: right foot; LF: left foot; T: torticollis; ED: embouchure dystonia
n. a.: not applicable; M: male, F: female; MRI: magnetic resonance imaging