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A systematic review of extensive music-practice as a risk factor for musician´s dystonia

Protocol

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Systematic review protocol: A systematic review on extensive music-practice as a pathogenetic factor in musician`s focal dystonia

1. Research Question

To assess the role of extensive music-practice as a risk factor for focal dystonia among professional musicians, and to examine any link between instrument-dependent demand on fine-motor skills and body part affected

2. Background

Musician`s dystonia is a task-specific movement disorder manifesting itself in professional musicians while playing an instrument¹. As there are no convincing therapeutic strategies available at present, disease onset usually terminates a professional musician`s career. According to current pathophysiologic concepts, musician`s dystonia is due to a constellation of intrinsic, i.e. genetic, and extrinsic, i.e. external circumstances and influences, factors². As musician`s dystonia often occurs after increasing practice time³, it has been hypothesized that extensive music-practice plays an important pathogenetic role. For writer`s cramp, a similar task-specific movement disorder, intensive writing was found to be a risk factor in a case-control study⁴. For musician`s dystonia, no systematic review of the literature has been conducted to date.

3. Aims

To assess the role of extensive music-practice as a risk factor for manifestation of focal dystonia among professional musicians and to examine any link between instrument-dependent demand on fine-motor skills and body part affected. The PICOS⁵ was defined as follows:

- Population: Professional musicians affected by focal dystonia
- Intervention/Exposure: extensive music-practice
- Comparison: general population (where applicable)
- Outcome: focal, task-specific dystonia while playing an instrument
- Study Design: any original research concerned with a group of patients

4. Search strategy

The following search terms will be used: (“musician” OR “performing artist” OR “pianist” OR “cellist” OR “drummer” OR “woodwind” OR “flutist” OR “violinist”) AND (“dystonia” OR “focal dystonia” or “musician`s dystonia” OR “occupational dystonia” OR “cramp” OR “embouchure”). In databases allowing only one search term, an abridged search strategy will be applied (“Musician`s dystonia”, MESH term). In German databases, an equivalent German search string will be developed.

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We will search multiple medical and music databases, i.e. EMBASE, MEDLINE, ISI Web of Science, PSYCINFO, BIOMED Central, Cochrane database, Library of the German Institute of Occupational Medicine, Library of the German Legal Occupational Health Institute (DGUV), BIOSIS Preview, WHO library, Computer-Assisted Information Retrieval System for Music (CAIRSS), British Library Sound Archive, Music Online, Wiley Online Library, Medical Problems of Performing Artists, Performing Arts Medicine Trust, Cambridge Companion Online, German Society for Music Physiology, Performing Arts Medicine Association, BIOMEDLIT and BIOMEDSEARCH.

We will also hand-search the reference lists of all included studies and relevant review articles, and will also search Google and Google Scholar.

Publication range will be set to 1950-2013, and English-language bias will be partly overcome by inclusion of German publications and consultation of interpreters in case of other publication languages.

5. Inclusion criteria and screening

Inclusion criteria:

- Original research article of any study design concerned with a group of patients
- Diagnosis of musician`s focal dystonia ascertained by a neurologist
- Report on clinical/epidemiologic first-hand patient information including body-part specific manifestation of dystonia and instrument(s) played

Exclusion criteria:

- Diagnosis performed by a physician other than a neurologist
- Animal experiments
- Non-empirical article
- Review
- Case reports (i.e. individual patient reports)

Screening of titles, abstracts and full text articles for eligibility will be undertaken by one reviewer, who will seek guidance from other reviewers in case of uncertainty. Publications by the same research groups drawing on the same or overlapping cohorts of patients will be assessed carefully, and only the more complete study will be included.

6. Data extraction

Data to be collected include the author, publication year and country, study design, number of affected musicians, instrument played in pre-specified instrument classes (i.e. keyboard, plucked string, bowed string, woodwind, brass, percussion), localisation of dystonic symptoms, sex, age at

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manifestation, concomitant diseases, family history and potential triggering factors, e.g. previous trauma, increase in practice intensity.

Data extraction of included studies will be performed by two independent reviewers, and any disagreements resolved through discussion.

7. Quality appraisal

The quality of included studies will be assessed using the Liverpool adaptation of the Newcastle-Ottawa-Scale⁶, enabling inclusion of a broad variety of study designs. According to this quality appraisal tool, 0-2 credit points can be attributed to each item. The quality domains examined are:

- Study design: Evidence of selection and response bias (yes: 0 credit points, maybe: 1 credit point; no: 2 credit points)
- Exposure assessment: Description of instrument played, quantification of practice time (no description: 0 credit points, vague description: 1 credit point; detailed description: 2 credit points), evidence of measurement bias (yes: 0 credit points, maybe: 1 credit point; no: 2 credit points)
- Outcome assessment: Diagnosis of dystonia (self-reported: 0 credit point; physician: 1 credit point; specialised physician: 2 credit points), evidence of ascertainment bias (yes: 0 credit points, maybe: 1 credit point; no: 2 credit points)
- Data analysis: Adjustment for confounding factors (no: 0 credit points; major factors accounted for: 1 credit point; detailed adjustment: 2 credit points)

A maximum of 16 credit points is possible, based on the sum of all credit points achieved in the above described quality domains; the higher the number of credit points, the higher the quality of the study.

Quality appraisal will be performed by two independent reviewers, and any disagreements will be resolved through discussion.

Data synthesis

Extracted data will be assembled in tables and/or graphically according to pre-specified instrument class, and a possible relationship between body-part specific manifestation of dystonic symptoms and instrument class will be examined.

Depending on data availability, sensitivity analyses will be conducted in relation to study design and possible determinants of musician's dystonia other than extensive music-practice, such as at manifestation, sex, concomitant diseases and triggering factors.

To assess whether the relationship between extensive music-practice and manifestation of focal dystonia in musicians is likely to be causal, the Bradford-Hill viewpoints^{7,8} will be applied. These are:

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strength of association, consistency, specificity, temporal sequence, biological gradient, plausibility, coherence, experiment and analogy.

9. Accounting for bias

We will attempt to minimise publication bias by searching across a broad range of electronic as well as grey literature database.

English-language bias will be minimised by inclusion of German publications and consultation of interpreters in case of other publication languages.

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Literature

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