Oral Dyskinesia: A Clinical Overview

Pierre J. Blanchet, MD, FRCP(C), PhD[a]/Pierre H. Rompré, MSc[b]/Gilles J. Lavigne, DMD, PhD, FRCD(C)[c]/Claude Lamarche, DMD, MSc[d]

Purpose: Dentists may be the first health care professionals to recognize unusual and abnormal oral movements collectively termed oral dyskinesias. The aims of this clinical overview are to raise the dental community's awareness about this important and complex topic and describe the clinical features and management of the main entities. ***Materials and Methods:*** A MEDLINE search of the different entities reported in the English and French literature was conducted. The main findings of a field study on oral dyskinesia were also reviewed. ***Results:*** Involuntary movement disorders are often drug related. In other cases, excessive oral movements may occur at any age in relation to various neuropsychiatric conditions. Orofacial dystonia apparently triggered by dental procedures has also been reported. Edentulousness has been associated with oral stereotypes. In a survey of 352 edentulous elderly individuals attending daycare centers, only 7% displayed visible oral sterotypes, and ill-fitting dentures were suggested as a possible triggering factor for the majority. ***Conclusion:*** A multidisciplinary evaluation is desirable in the care of individuals with oral dyskinesia and in the selection of those who may benefit from a prosthodontic approach. A good knowledge of potentially offending drugs may allow avoidance of unnecessary procedures. *Int J Prosthodont 2005;18:10–19.*

Oral dyskinesias consist of abnormal, involuntary, uncontrollable movements predominantly affecting the tongue, lips, and jaw. They often vary in complexity, distribution, and severity. They may go unnoticed or cause social embarrassment, oral traumatic injury, speech difficulty, chewing and eating disorders, inability to wear prosthetic devices, or affect professional activities (eg, embouchure dystonia in musicians).

[a]*Associate Professor, Department of Stomatology, Faculty of Dental Medicine, University of Montreal; and Department of Medicine, University of Montreal Hospital Centre, Canada.*
[b]*Research Assistant, Department of Oral Health, Faculty of Dental Medicine, University of Montreal, Canada.*
[c]*Professor, Department of Oral Health, Faculty of Dental Medicine, University of Montreal, Canada.*
[d]*Professor, Department of Restorative Dentistry, Faculty of Dental Medicine, University of Montreal, Canada.*

Correspondence to: *Dr Pierre J. Blanchet, Andre-Barbeau Movement Disorders Unit, Universite de Montreal Hospital Centre (CHUM), Room 7-561, 3840 Saint-Urbain, Montreal, Quebec, H2W 1T8 Canada. Fax: + (514) 412-7139. e-mail: pierre.j.blanchet@umontreal.ca*

Interest in oral dyskinesias has grown tremendously in the last 50 years with the advent of the conventional ("typical") antipsychotic drugs. Because the diagnosis essentially relies on clinical assessment, recognition and proper management are not always straightforward. Any case of oral dyskinesia should be initially considered drug induced until proven otherwise. This allows a proper investigation of the patient drug history as a part of the medical history before other dental or medical interventions are proposed. Neurologic and peripheral orodental factors contribute to a variable extent depending on the cause, suggesting that a multidisciplinary evaluation may be preferable to improve patient management.

This clinical overview attempts to raise the awareness of the dental community about this important and complex topic and cover the features and management of the main clinical entities seen in adulthood, as described in the English and French literature gathered following a MEDLINE search. The authors' own data and views on edentulous dyskinesia are also presented.

Table 1 Differential Diagnosis of Oral Dyskinesias

Subtype	Cause
Bucco-linguo-mandibular dyskinesia (often complex)	Drug induced Neurodegenerative disorders Huntington's disease neuroacanthocytosis Hepatocerebral degeneration Chronic schizophrenia
Oral dyskinesia (often mild and stereotyped)	Drug induced Subcortical infarcts Neuropsychiatric conditions chronic schizophrenia autism Rett syndrome Dementia Mental retardation Peripherally induced edentulous dyskinesia denture-related dyskinesia (ill-fitting prosthesis) Spontaneous (detailed characterization lacking)
Oromandibular dystonia (jaw-closing, jaw-opening, lateral jaw deviation)	Drug induced (tardive dystonia) Idiopathic (sporadic, inherited) associated with blepharospasm (Meige syndrome) Neurodegenerative disorders Huntington's disease neuroacanthocytosis (with "eating dystonia") Wilson's disease Secondary basal ganglia/rostral brain stem disorders head trauma subcortical infarcts infections tumors metabolic conditions Peripherally induced trauma dental interventions
Diurnal bruxism (teeth clenching or grinding)	Drug induced (antipsychotics, antidepressants) Acute toxin exposure (ecstasy, amphetamines) Associated with dystonia Subcortical infarcts Oculomasticatory myorhythmia (Whipple's disease)
Tics	Drug induced Idiopathic Gilles de la Tourette syndrome Chronic motor tics Head trauma Intoxication (CO, gasoline) Infections/postinfectious Hallervorden-Spatz syndrome
Perioral tremor ("rabbit syndrome")	Drug induced (antipsychotics) Parkinson's disease

Oral Dyskinesias

General Description

Oral dyskinesias are classified according to the phenomenology of the abnormal movements or cause (Table 1). The abnormal movements are often brief, recurrent, and more or less patterned and coordinated to produce lip pursing, pouting, smacking and sucking, licking, tongue writhing and protrusion, and chewing. These movements may be described as "stereotyped" if they are highly patterned and predictable. A complex symptomatology may be observed to justify the classic label of "bucco-linguo-masticatory dyskinesia," typical of chronic antipsychotic drug exposure. The dyskinetic movements may be restricted to the oral region or extend to trunk and limb musculature, particularly in drug-induced and complex neurologic cases.

Oral dyskinetic movements, albeit repetitive, are usually not as rhythmic as tremor. One exception is bruxism. Oral dyskinesia must also be distinguished from "rabbit syndrome," a form of slow, vertical, labial tremor sparing the tongue and resembling the rhythmic chewing movements of a rabbit. Rabbit syndrome is seen in fewer than 5% of those on conventional antipsychotic drugs.[1–3] Oral dyskinesia may resemble tics, which are

brief, sudden, irregularly occurring, purposeless, repetitive movements or sounds, most often starting in childhood. These tend to wax and wane, may be produced as a response to an involuntary urge (described as a sensation of tingling, itching, or aching), are commonly accompanied by vocal manifestations, and are distinctly suppressible for variable periods. Voluntary control is limited by an unpleasant inner tension buildup pressing the individual to release the tics.

The complications resulting from oral dyskinesia are likely to be first diagnosed by the dentist and include the following:

- Tooth wear
- Tooth and denture damage
- Accelerated bone loss in edentulous patients
- Oral pain
- Temporomandibular joint (TMJ) degeneration
- Mandibular luxation
- Friction/biting injuries (tongue, cheek bites)
- Speech impairment
- Dysphagia
- Chewing difficulties
- Inadequate food intake and weight loss
- Displacement/impaired retention of removable dental prostheses (reduced tolerance)
- Social embarrassment (unemployment, isolation, depression)

Oral dyskinesia may be an under-recognized source of pain[4,5] that has multiple potential causes: muscle stiffness, TMJ degeneration,[6] mandibular luxation, mucosal and gingival traumatic lesions, or impaired retention with displacement of dental prosthetic devices. Chewing difficulty, related to the dyskinetic movements or inability to wear dentures, and dysphagia may occur. Speech impairment is observed in a fraction of cases, adding to the social embarrassment some patients experience for cosmetic reasons.

Subtypes of Oral Dyskinesia

Tardive dyskinesia. Delayed involuntary movements occurring during chronic antipsychotic drug therapy were recorded in the late 1950s, but the term "tardive dyskinesia" (TD) was not introduced until 1964.[7] The term encompasses several types of abnormal involuntary movements, including stereotypes, dystonia, and chorea, which may coexist in the same patient or occur in isolation or with a dominant subtype referred to as tardive stereotypes, tardive dystonia, and so forth. Typically, tardive stereotypes predominantly affect the labial, lingual, and mandibular musculature to a variable degree (hence the term "bucco-linguo-masticatory dyskinesia"), but frequent involvement of the limb musculature helps distinguish drug-induced cases from other conditions.

The prevalence of TD among patients chronically treated with conventional antipsychotic drugs (Table 2), all potent dopamine D2 receptor blockers, is variable in the literature, with an average estimate of 20%.[8] The incidence in young adults is about 4% to 5% per year.[9] However, the risk of drug-induced dyskinesia is reported to be three to six times greater in the elderly population in comparison to younger individuals.[10,11] Accordingly, the minimum length of antipsychotic drug exposure required to propose a causal relation between the dyskinesia and the offending drug, which is typically at least 3 months in younger individuals, can be as short as 1 month in elderly people. Complex pharmacokinetic and pharmacodynamic factors may explain this differential susceptibility.

The possibility of a genetic vulnerability with associated polymorphisms has been suggested for the drug-metabolizing enzyme CYP2D6[12,13] and the dopamine D3 receptor gene.[14] The use of high-potency conventional antipsychotics, or intramuscular, slow-release preparations for noncompliant patients, also carries a higher risk of TD (Table 2) compared to second-generation (so-called atypical) antipsychotics, which display lower affinity for dopamine D2 receptors and higher affinity for serotonergic 5-HT_2 receptors.[15,16] However, the TD risk entailed by some of the atypical drugs is felt by some investigators to remain significant.[17] The course of TD is uncertain and unpredictable, and rates of persistent remission ranging between 30% and 50% have been reported.[18,19] TD may improve over the years at the expense of worsening of parkinsonism in psychiatric patients who remain on antipsychotic drug treatment.[20] Longstanding TD is less likely to remit spontaneously.

The pathophysiology of TD is not well understood, but striatal dopamine D2 receptor supersensitivity has been the traditional and oversimplistic explanation.[21] Receptor supersensitivity is expected to take place early and in virtually all treated patients, and to disappear following drug withdrawal, whereas TD may appear late, occurs in a fraction of patients, and often persists indefinitely after drug withdrawal. Other transmitter systems (gamma-amino butyric acid [GABA], glutamate, opioid peptides) are likely involved. Some data also suggest that peripheral orodental factors contribute to the expression of TD: *(1)* orofacial motor ratings were more severe in edentulous subjects in one study,[22] but results comparable to dentate subjects have also been obtained[23]; and *(2)* dental prosthetic treatment has been shown to ameliorate TD.[24] Further prospective studies with attempts to limit selection and observer bias are warranted to clarify this issue.

Table 2 Main Offending Drugs Associated with Oral Dyskinesia

Generic name	Trade name
Conventional antipsychotics	
Chlorpromazine	Largactil, Thorazine
Fluphenazine	Moditen, Modecate, Prolixin, Permitil
Haloperidol	Haldol
Levopromazine	Nozinan
Perphenazine	Trilafon, Trilifan
Pimozide	Orap
Prochlorperazine	Compazine
Thioridazine	Mellaril, Melleril
Thioproperazine	Majeptil
Thiothixene	Navane
Trifluoperazine	Stelazine
Atypical antipsychotics	
Clozapine	Clozaril, Leponex
Olanzapine	Zyprexa
Quetiapine	Seroquel
Risperidone	Risperdal
Antiemetics	
Metoclopramide	Reglan, Maxeran, Primperan
Prochlorperazine	Stemetil
Promethazine	Phenergan
Antiparkinsonian agents	
Levodopa	Prolopa, Madopar, Modopar, Sinemet, Sinemet-CR
Benztropine	Cogentin
Procyclidine	Kemadrin
Trihexiphenidyl	Artane, Parkinane
Tricyclic antidepressants	
Amitryptiline	Elavil, Laroxyl
Doxepine	Sinequan, Quitaxon
Imipramine	Tofranil
Selective serotonin reuptake inhibitors (SSRI)	
Fluoxetine	Prozac
Fluvoxamine	Luvox, Floxyfral
Paroxetine	Paxil, Deroxat
Sertraline	Zoloft
Lithium (usually in toxic range)	Carbolith, Duralith, Lithane, Teralithe
Anticonvulsants (especially with coexisting brain lesions)	
Diphenylhydantoin	Dilantin, Di-Hydan
Carbamazepine	Tegretol
Primidone	Mysoline
Phenobarbital	Gardenal, Alepsal
Antihistamines (H1 or H2 receptor blockers)	
Diphenhydramine	Benadryl
Cimetidine	Tagamet
Ranitidine	Zantac, Azantac, Raniplex
Oral contraceptives (similar to chorea gravidarum)	

Clinicians should be aware that other drugs and conditions can cause oral dyskinesias and bruxism (ie, teeth clenching or grinding). The main drug offenders are hidden dopamine receptor–blocking agents known for their antiemetic properties (Table 2). TD-like disorders can be triggered by tricyclic antidepressants[25,26] and selective serotonin reuptake inhibitors (SSRI).[27–30] The latter drug class can also produce bruxism.[31] Other offending medications associated with oral dyskinesia include lithium, antiparkinsonian medications, anticonvulsants (particularly with coexisting brain lesions), and histamine H2 receptor blockers. Rarely, cocaine can cause transient dyskinesias, or "crack dancing."[32] Acute intoxication with amphetamine and 3,4-methylenedioxymethamphetamine (MDMA, or ecstasy) has been associated with bruxism and tooth wear.[33–36] These toxins acutely enhance the brain release of dopamine and/or serotonin from nerve endings.

Edentulous dyskinesia. Edentulousness is thought to be a common cause of oral dyskinesia, although the number of studies on this association is surprisingly low. One study[37] compared 75 consecutive edentulous subjects (74 men, 1 woman; mean age 62 years) seen in a Veterans Administration dental clinic to 75 age-matched controls (37 men, 38 women) with natural teeth. Tooth extraction had been performed more than a decade before in the former group. Twelve (16%) of the edentulous subjects had oral dyskinesia, whereas none of the control subjects were affected. The stereotypes were mild in 9 subjects and marked in the other 3. Stereotyped smacking and pursing of the lips, lateral deviation and protrusion of the tongue, and occasionally lateral deviation and protrusion of the jaw have been documented. Unlike drug-induced dyskinesia, the movements observed were always confined to the oral region and never dystonic, and no tongue movements were recorded when the mouth was open. It is worth noting that 6 of the 12 edentulous subjects with dyskinesia wore no dentures, and 1 additional patient had ill-fitting dentures. Ten of these 12 subjects were aware of the presence of oral movements. Pain was not a feature. Differences in alcohol intake and smoking habits were not controlled between groups.

A recent field study on oral dyskinesia observed non-institutionalized frail individuals attending daycare centers, at least 60 years of age, and asked them to fill out a brief survey concerning their medical and dental history.[5] The main objective was to determine the prevalence of and isolate relevant orodental factors for oral dyskinesia. A total of 1,018 subjects (69.3% women) were observed during outpatient activities (exercise classes, workshops, social activities, seminars). Thirty-eight subjects (29 women, 9 men) were thought to display oral dyskinesia of no apparent cause, for an overall prevalence rate of 3.7% (95% confidence interval 2.6 to 4.9), 4.1% for women and 2.9% for men. In the surveys filled out by 508 volunteers, the subjects with oral dyskinesia of no apparent cause reported more frequent ill-fitting dental devices, oral pain, and a lower rate of perception of good oral health compared to nondyskinetics. In these subjects, oral dyskinesia generally consisted of mild stereotyped masticatory or labial move-

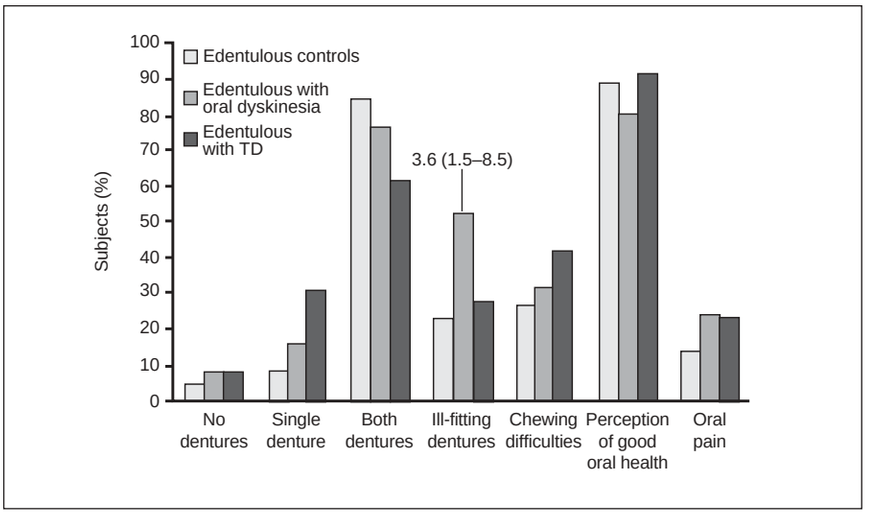

Fig 1 Survey answers of edentulous subjects concerning the orodental condition: controls without oral dyskinesia (n = 307), subjects with suspected spontaneous oral dyskinesia (n = 25), and subjects with probable antipsychotic drug–induced dyskinesia (TD; n = 13). Odds ratio (95% confidence interval) is provided where appropriate ($P < .010$).

ments, compared to the more complex phenomenology of probable drug-induced cases, and appeared to match previous descriptions.[37] The response profile of 352 edentulous subjects (69% of responders) could be further analyzed: 307 of these subjects (87%) showed no apparent oral dyskinesias, 25 (7%) displayed oral dyskinesia that could not be readily attributed to a secondary cause, 13 (4%) had probable TD, and 7 (2%) were thought to have antidepressant drug–related dyskinesia. The proportion of edentulous subjects not wearing any dentures was low (5.7% in the surveyed population). However, 52% of the edentulous subjects with oral dyskinesia of no apparent cause reported problems with ill-fitting dentures, a proportion substantially higher than in the nondyskinetic subjects ($P = .005$; odds ratio 3.6, 95% confidence interval 1.5 to 8.5) or those with TD (Fig 1). No other distinct orodental factor was identified. It was concluded that "spontaneous" oral dyskinesia is comparatively infrequent in the elderly population in relation to those exposed to antipsychotic drugs. The authors suggest that certain orodental factors (eg, ill-fitting dentures, oral discomfort), in addition to edentulousness, may trigger oral dyskinesia. Further studies are underway to clarify these issues. Cases of edentulous dyskinesia and denture-related dyskinesia could well be amenable to correction using a careful prosthodontic approach, as described below.

Spontaneous oral dyskinesia. Early descriptions made prior to the introduction of antipsychotic drugs have included comments about the presence of lip smacking, chewing, and choreoathetoid limb movements in patients with chronic schizophrenia.[38,39] A review article estimates the prevalence rate of dyskinesia in never-treated individuals with chronic schizophrenia to be 12%.[40] Another study of 108 never-treated patients, 37 of whom were reexamined at 18 months, emphasizes the fluctuating course of dyskinesia in these individuals and reported a prevalence rate of 57%.[41] In one study comparing the motor ratings of institutionalized patients with schizophrenia chronically exposed (n = 364; mean age 56.9 ± 13.3 years) or never exposed (n = 47; mean age 66.7 ± 11.7 years) to antipsychotic drugs, a striking similarity was observed in terms of prevalence, severity, and body distribution of the involuntary movement pattern.[42] Twenty-five of the 47 antipsychotic-free subjects showed evidence of dyskinesias, mainly in the oromandibular region, supporting the claim that antipsychotic drugs are innocent bystanders in the development of dyskinesia in people with schizophrenia. Nevertheless, some differences between the two groups were reported: Patients exposed to antipsychotics showed more severe lingual dyskinesia and grimacing, more frequent upper limb dyskinesia, and less frequent head nodding than antipsychotic-free subjects. Dystonic features were uncommon. The dental status of the subjects was not reported.

Other data suggest that the net contribution of antipsychotic drugs to dyskinesia induction in people with schizophrenia cannot be rejected on clinical grounds. In one study comparing psychiatric patients and normal subjects, abnormal choreiform limb movements were almost restricted to antipsychotic-treated patients.[43]

Antipsychotic drugs and other dopamine receptor–blocking agents may cause persistent dyskinesias in nonpsychiatric patients (see above), as well as in nonhuman primates. Finally, antipsychotic drugs can produce distinct tardive movement disorders such as dystonia and tremor, which do not arise spontaneously. Better knowledge of the complex biochemical and molecular changes triggered in the brain by antipsychotic drugs will further understanding of how TD emerges and how to manage it more effectively.

While the concept of spontaneous dyskinesias in patients with chronic schizophrenia is not disputed, it seems nonspecific and extends to other brain conditions such as autism, mental retardation, and Rett syndrome. In Alzheimer's disease, reported estimates of the incidence of orofacial dyskinesia in antipsychotic-free patients have varied between 10% and 40%.[44–46] In another report, only 1% of 500 demented, antipsychotic-free, elderly patients displayed dyskinesias.[47] In these patients, brief exposure to an antipsychotic medication or any other offending drug, more likely to be overlooked, may be sufficient to trigger dyskinesia and inflate prevalence figures. The impact of institutionalization and resulting deprivation has been little addressed as a risk factor.[48]

The question of the occurrence of spontaneous orofacial dyskinesia in normal elderly individuals has been addressed to determine if the aging process alone is sufficient to trigger it. The prevalence estimates have greatly varied depending on the population studied. Prevalence figures as high as 18% (38 of 211 individuals)[49] and 31.7% (120 of 378 individuals)[50] were reported, but these subjects were institutionalized and the presence of dementia was not assessed. In 29 individuals at least 50 years of age, 20% showed signs of orofacial dyskinesia,[43] but other estimates have been much more conservative, in the range of 1% to 4%.[47,51,52] Dyskinesias are generally mild and almost restricted to the oral region. Women seem more frequently affected. The orodental condition of these subjects was not reported. It is tempting to speculate that at least a fraction of these cases consisted of edentulous dyskinesia or denture-related dyskinesia.

Oromandibular Dystonia

General Description

Oromandibular dystonia is produced by involuntary, excessive, and sustained muscle contractions involving the mouth, jaw, and tongue, causing painful spasms, twisting, and jerky movements. Lip retraction and grimacing, tongue rolling interfering with speech and food intake (as food is pushed out of the mouth or difficult to swallow), involuntary jaw closure with trismus, and jaw opening and lateral deviation may be observed. Meige syndrome is diagnosed if dystonic movements of the upper face coexist. Unlike other movement disorders, sensory signals ("geste antagoniste," or "sensory trick") provided by holding a seed or candy in the mouth, placing a straw or piece of plastic between the rear molars on one side, or a light touch on a tooth or chin may reduce the intensity of the dystonic spasms.[53,54] Dystonia is one of the most difficult movement disorders to characterize and is commonly misattributed to a psychogenic cause. Involuntary activity in oromandibular muscles causing teeth grinding or clenching, characteristic of idiopathic sleep bruxism, may also be observed as a daytime manifestation of oral dystonia and can also occur following acute or chronic antipsychotic drug exposure.[55] Oral dystonia is often more disabling than typical TD, and jaw movements may be so forceful as to cause tooth wear and loss. Food intake may be impaired because of jaw spasms or prominent lingual dystonia pushing food out of the oral cavity. In one case of orofacial dystonia seen by one of the authors, rupture of the lingual frenum with hematoma and swelling required urgent evaluation.

Subtypes of Oromandibular Dystonia

Tardive oral dystonia. Focal or generalized dystonia may develop in fewer than 5% of subjects exposed to antipsychotic drugs, typically after years of treatment.[56] This persistent condition must be distinguished from transient dystonic reactions occurring acutely following initiation of various dopamine receptor–blocking agents. The craniocervical region is the most commonly affected site.[57] The phenomenology is indistinguishable from that of idiopathic "primary" dystonia. Tardive dystonia may coexist with TD in a given patient. The spontaneous remission rate appears three- to fivefold lower than for typical TD.

Idiopathic "primary" dystonia. Sporadic cases of oral dystonia starting in adulthood occur as an isolated disorder (without additional neurologic features), much like other forms of focal dystonia such as spasmodic torticollis, essential blepharospasm, or writer's cramp. For some authors, these cases represent the category of "spontaneous" orofacial dyskinesia described in the literature outside the contexts of schizophrenia, cognitive impairment, and edentulousness.[58,59] The pathophysiology of dystonia is not well understood, but the physiologic inhibitory control of the basal ganglia of the forebrain over the thalamus and brain stem is thought to be defective.

Dental procedure–related dystonia. In rare case reports, disruption of the orofacial neuromuscular control has been implicated as a trigger for oromandibular dystonia. Sankhla et al[60] report on 27 cases (18 women) who had experienced at least one dental procedure (22 cases), facial or orodental trauma (4 cases), or frontal sinus obliteration (1 case) prior to the onset of oromandibular dystonia. Four patients with ill-fitting dentures and 1 with an ill-fitting fixed partial denture "gave a history of trying to adjust their bite by manipulating the jaw to adapt to the newly fitted dentures or the bridge." The mean latency between the insult and onset of the movement disorder was 65 days (range 1 to 365 days), but 12 patients had the onset of their clinical manifestations of dystonia within 1 week. The mean age of onset was 50 years (range 16 to 73 years), with a peak between 50 and 60 years of age. Spreading of the manifestations of oromandibular dystonia to other muscle groups occurred in some cases. One subject had preexisting dystonic manifestations in her neck for 1 year, with spreading to the oromandibular region within 1 week after root canal treatment. Associated nocturnal bruxism (in 4 cases) and diurnal bruxism (in 2 of the 4 cases) also developed but was somewhat less frequent than in a control group with idiopathic oromandibular dystonia. Pain was more frequently reported in the peripherally induced traumatic cases compared to the control idiopathic group, but the difference did not reach significance. Patients of both groups used sensory tricks to reduce the oromandibular manifestations.

Schrag et al[61] describe eight subjects with natural dentition, 35 to 59 years of age, in whom jaw dystonia occurred within days to a few weeks after a dental procedure (tooth extraction in five, fixed partial denture insertion and several fillings in one, unspecified in two), often spreading to other muscle groups (tongue, palate, lips, face, neck, or hand) thereafter. The onset of the atypical jaw-deviating spasms in five of these cases, all women, was associated with persistent jaw pain and dysesthesia. None of these patients had ever taken antipsychotic drugs, showed focal brain lesions, or reported a family history of dystonia. No direct relation was found between the severity of the injury and the development of dystonia. A coincidence or unmasking effect by the dental procedure of a latent movement disorder cannot be entirely ruled out, nor can a psychogenic disorder. In view of the scarcity of such cases, additional coexisting factors may need to be present for the dystonia to evolve. Thus, definite conclusions about the strength of the causal relation between dental procedures and dystonia should not be drawn prematurely, as such cases are uncontrolled and putative etiologies based on clinical opinion only, making the interpretations potentially biased by multiple confounding variables.

Daytime bruxism. Cases of dystonia or diurnal bruxism (teeth clenching and grinding) have also been described in the context of drug and toxin exposure[31] and various neurologic conditions, including head trauma, subcortical ischemic brain lesions affecting basal ganglia structures,[62] petroclival meningioma, encephalitis, syphilis, and metabolic conditions. A distinct form of persistent (diurnal and nocturnal) bruxism can arise as a complication of Whipple's disease (systemic *Tropheryma whipplei* infection), producing a picture of "oculomasticatory myorhythmia" or "oculofacio-skeletal myorhythmia," with rhythmic convergent eye oscillations and synchronous contractions of facial, jaw, and other muscles.[63] These cases must be diagnosed and treated early with antibiotics to minimize neurologic sequelae.

Management of Oral Dyskinesia

The emphasis of management strategies needs to be directed toward prevention rather than palliative treatment in drug-induced oral dyskinesia.[21] Indeed, TD carries the potential to become persistent even though the offending drug is stopped. Thus, the early detection of vermicular tongue movements by the dentist should be immediately reported to the treating physician. In clinically significant TD, withdrawal of the offending drug should be contemplated. This is not always possible in the psychiatric patient population. Switching to an "atypical" antipsychotic drug would be the next option. If TD persists, palliative treatment with tetrabenazine, a central monoamine depleter not causing TD by itself, may reduce its intensity.[64] This drug can be used alone or in combination with the benzodiazepine clonazepam, which may increase somnolence and carries a risk of dependence and tachyphylaxis over time. Dystonic cases may benefit from a trial of anticholinergic drug, and clonazepam and baclofen have had some success. For oromandibular dystonia or bruxism, intramuscular botulinum toxin is an excellent option.[65–68] Selected disabled and refractory cases may respond to a neurosurgical intervention.[69–71]

The dentist or prosthodontist is often a key member in the management of oral dyskinesia. First, orodental complications must be evaluated and treated diligently. Replacing ill-fitting dentures or offering proper fit of a single denture on the supporting structures can preclude ulcers and discomfort. For cases of jaw-closing dystonia clearly responsive to a sensory trick, a simple bite-raising soft device fitting between the molars to prevent jaw closure by a few millimeters can improve function.[53,54] In one example, a ball clasp between the molars secured the device, which extended onto the lingual tooth surface for better retention.[54] Such devices should be introduced early to prevent tooth wear and

TMJ dysfunction. In edentulous subjects wearing dentures, proper adjustment of the occlusion and relining have also successfully improved the intensity of oral dyskinesia, drug induced or not.[72–76] The final vertical dimension selected was comparatively greater than that of the previous denture in several reported cases.[72,73,76]

An unconventional prosthetic therapy, positioning the mandible anteriorly in occlusion, has been developed by Sutcher et al.[72] Between 1950 and 1970, they examined many edentulous "difficult denture" patients who exhibited "wavering, uncertain patterns of mandibular movement, which were frequently accompanied by involuntary, irregular movements of the tongue, lips, and chin," resembling dystonia and interfering with voluntary motor control. In some cases, symptoms were felt to be reduced after "proper" construction of an occlusal prosthesis (a modified dental splint) and complete dentures restoring physiologic craniomandibular relationships were made. In a detailed account of four subjects (three women, one man) aged between 45 and 72 years, all had worn dentures for many years (complete maxillary and mandibular dentures in three, a complete maxillary over a removable partial mandibular denture in another case) and been found to have grossly incorrect denture occlusions. They displayed uncontrollable "jumping" of the jaw; involuntary dystonic movements of oral, facial, and cervical muscles, with tongue tremor and protrusion; severe jaw-opening dystonia, with constant dystonic movements of the tongue; and symptoms of orofacial dyskinesia unrelated to any history of drug treatment. Onset was spontaneous and gradual in three cases and occurred several months after acquiring a new set of uncomfortable, ill-fitting dentures in another patient. These patients displayed a typical geste antagoniste, keeping a hand or finger on the side of the mandible or under the chin to counteract dystonia. Incorrect occlusion in these patients was improved by adding a layer of acrylic resin over the teeth of the maxillary denture, then providing the strength needed to help patients approximate their jaws and achieve a bite lock, leaving a groove in the occlusal prosthesis into which the mandibular denture could fit to restrict the involuntary mandibular movements. The labial portion of the mandibular denture was almost entirely covered by the occlusal prosthesis, which was 20 mm deep. Further individual occlusal modifications were proposed as needed. Sustained improvement in dyskinetic manifestations was observed, taking place almost instantaneously or within hours. During follow-up, the locking feature of the prosthesis was progressively reduced to almost a flat plane as involuntary movements decreased, and the prosthesis was then replaced by a new set of dentures providing correct occlusion.

Sutcher et al[72] argue that edentulousness (along with incorrect occlusions produced by inadequate dentures) chronically distorts most of the peripheral proprioceptive input from the stomatognathic system necessary for central sensorimotor integration, thereby promoting dyskinesia. This hypothesis alone does not account for the delay between the edentulousness and onset of the abnormal movements or the seemingly low frequency of oral dyskinesia in edentulous subjects. Nevertheless, the efficacy of the proposed occlusal therapy exempliqfies the contribution of sensory inputs to the expression of dystonia. The difficulty with this approach lies in the fact that it is an individualized and somewhat arbitrary method of positioning the mandible anteriorly in occlusion. It is unclear whether the four cases described represent the entire patient population undergoing treatment, or whether they display a homogeneous or heterogeneous disorder. The benefit of Sutcher et al's approach has been reported anecdotally in few cases, and the difference in outcome between patients who do or do not display an effective geste antagoniste remains unclear. Nevertheless, this noncontrolled, empiric, peripheral sensory approach certainly warrants further attention.

Because denture retention may be troublesome in the presence of oral dyskinesia, an implant-stabilized removable mandibular prothesis has been proposed. Only a few isolated cases have been reported.[77,78] Ideally, this should not be attempted without prior discussion with the treating physician. Trauma to the implant site with local granulomatous tissue reaction may occur in relation to the dyskinetic mandibular movements and the wearing of a maxillary denture.[78]

Conclusion

Oral dyskinesia is variable in phenomenology, topography, severity, and functional impairment. It may be drug related or a sign of a neurologic condition at any age, or caused by orodental factors (edentulous dyskinesia, denture-related dyskinesia). Upon recognition of the dyskinesia, the dentist or prosthodontist should first report to the medical team primarily responsible for the diagnosis and management of the condition. It is desirable that any attempt to correct the denture occlusion and provide a more balanced craniomandibular relationship be discussed and agreed upon to avoid unnecessary and contradictory approaches. Any team approach in oral dyskinesia management should include an interested general dentist or prosthodontist.

Acknowledgments

This work was supported by the Canadian Institutes of Health Research and the Quebec Health Research Fund (FRSQ). Mrs Marie T. Parent provided assistance during the preparation of the manuscript.

References

1. Villeneuve A. The rabbit syndrome: A peculiar extrapyramidal reaction. Can Psychiatr Assoc J 1972;17(suppl 2):SS69–SS72.
2. Yassa R, Lal S. Prevalence of the rabbit syndrome. Am J Psychiatry 1986;143:656–657.
3. Schwartz M, Hocherman S. Antipsychotic-induced rabbit syndrome: Epidemiology, management and pathophysiology. CNS Drugs 2004;18:213–220.
4. Bassett A, Remick RA, Blasberg B. Tardive dyskinesia: An unrecognized cause of orofacial pain. Oral Surg Oral Med Oral Pathol 1986;61:570–572.
5. Blanchet PJ, Abdillahi O, Beauvais C, Rompré PH, Lavigne GJ. Prevalence of spontaneous oral dyskinesia in the elderly: A reappraisal. Mov Disord 2004;19:892–896.
6. Osborne TE, Grace EG, Schwartz MK. Severe degenerative changes of the temporomandibular joint secondary to the effects of tardive dyskinesia: A literature review and case report. J Craniomandib Pract 1989;7:58–62.
7. Faurbye A, Rasch PJ, Petersen PB, Brandborg G, Pakkenberg H. Neurological symptoms in pharmacotherapy of psychoses. Acta Psychiatr Scand 1964;40:10–27.
8. Casey DE. Neuroleptic-induced extrapyramidal syndromes and tardive dyskinesia. In: Hirsch S, Weinberger DR (eds). Schizophrenia. Oxford, UK: Blackwell, 1995:546–565.
9. Kane JM, Woerner M, Lieberman J. Tardive dyskinesia: Prevalence, incidence, and risk factors. J Clin Psychopharmacol 1988;8(4 suppl):52S–56S.
10. Woerner MG, Alvir JMJ, Saltz BL, Lieberman JA, Kane JM. Prospective study of tardive dyskinesia in the elderly: Rates and risk factors. Am J Psychiatry 1998;155:1521–1528.
11. Jeste DV. Tardive dyskinesia in older patients. J Clin Psychiatry 2000;61(suppl 4):27–32.
12. Kapitany T, Meszaros K, Lenzinger E, et al. Genetic polymorphisms for drug metabolism (CYP2D6) and tardive dyskinesia in schizophrenia. Schizophr Res 1998;32:101–106.
13. Lam LC, Garcia-Barcelo MM, Ungvari GS, et al. Cytochrome P450 2D6 genotyping and association with tardive dyskinesia in Chinese schizophrenic patients. Pharmacopsychiatry 2001;34:238–241.
14. Lerer B, Segman RH, Fangerau H, et al. Pharmacogenetics of tardive dyskinesia: Combined analysis of 780 patients supports association with dopamine D3 receptor gene Ser9Gly polymorphism. Neuropsychopharmacology 2002;27:105–119.
15. Casey DE. Will the new antipsychotics bring hope of reducing the risk of developing extrapyramidal syndromes and tardive dyskinesia? Int Clin Psychopharmacol 1997;12(suppl 1):S19–S27.
16. Kane JM. Extrapyramidal side effects are unacceptable. Eur Neuropsychopharmacol 2001;11(suppl 4):S397–S403.
17. Halliday J, Farrington S, MacDonald S, MacEwan T, Sharkey V, McCreadie R. Nithsdale Schizophrenia Surveys 23: Movement disorders. Br J Psychiatry 2002;181:422–427.
18. Barnes TRE, Kidger T, Gore SM. Tardive dyskinesia: A 3-year follow-up study. Psychol Med 1983;13:71–81.
19. Kane JM, Woerner M, Borenstein M, Wegner J, Lieberman J. Integrating incidence and prevalence of tardive dyskinesia. Psychopharmacol Bull 1986;22:254–258.
20. Fernandez HH, Krupp B, Friedman JH. The course of tardive dyskinesia and parkinsonism in psychiatric inpatients: 14-year follow-up. Neurology 2001;56:805–807.
21. Blanchet PJ. Antipsychotic drug-induced movement disorders. Can J Neurol Sci 2003;30(suppl 1):S101–S107.
22. Myers DE, Schooler NR, Zullo TG, Levin H. A retrospective study of the effects of edentulism on the severity rating of tardive dyskinesia. J Prosthet Dent 1993;69:578–581.
23. Sandyk R, Kay SR. Edentulousness and neuroleptic-induced neck and trunk dyskinesia. Funct Neurol 1990;5:361–363.
24. Sutcher H, Soderstrom J, Perry R, Das A. Tardive dyskinesia: Dental prosthetic therapy. Panminerva Med 1998;40:154–156.
25. Fann WE, Sullivan JL, Richman BW. Dyskinesias associated with tricyclic antidepressants. Br J Psychiatry 1976;128:490–493.
26. Vandel P, Bonin B, Leveque E, Sechter D, Bizouard P. Tricyclic antidepressant-induced extrapyramidal side effects. Eur Neuropsychopharmacol 1997;7:207–212.
27. Ellison JM, Stanziani P. SSRI-associated nocturnal bruxism in four patients. J Clin Psychiatry 1993;54:432–434.
28. Leo RJ. Movement disorders associated with the serotonin selective reuptake inhibitors. J Clin Psychiatry 1996;57:449–454.
29. Gerber PE, Lynd LD. Selective serotonin-reuptake inhibitor-induced movement disorders. Ann Pharmacother 1998;32:692–698.
30. Bostwick JM, Jaffee MS. Buspirone as an antidote to SSRI-induced bruxism in 4 cases. J Clin Psychiatry 1999;60:857–860.
31. Winocur E, Gavish A, Voikovitch M, Emodi-Perlman A, Eli I. Drugs and bruxism: A critical review. J Orofac Pain 2003;17:99–111.
32. Daras M, Koppel BS, Atos-Radzion E. Cocaine-induced choreoathetoid movements ("crack dancing"). Neurology 1994;44:751–752.
33. Ashcroft GW, Eccleston D, Waddell JL. Recognition of amphetamine addicts [letter]. Br Med J 1965;1:57.
34. Peroutka SJ, Newman H, Harris H. Subjective effects of 3,4-methylenedioxymethamphetamine in recreational users. Neuropsychopharmacology 1988;1:273–277.
35. Milosevic A, Agrawal N, Redfearn PJ, Mair LH. The occurrence of toothwear in users of ecstasy (3,4 methylenedioxymethamphetamine). Community Dent Oral Epidemiol 1999;27:283–287.
36. Nixon PJ, Youngson CC, Beese A. Tooth surface loss: Does recreational drug use contribute? Clin Oral Invest 2002;6:128–130.
37. Koller WC. Edentulous orodyskinesia. Ann Neurol 1983;13:97–99.
38. Jeste DV, Wyatt RJ. Changing epidemiology of tardive dyskinesia: An overview. Am J Psychiatry 1981;138:297–309.
39. Villeneuve A, Turcotte J, Bouchard M, Côté JM, Jus A. Release phenomena and iterative activities in psychiatric geriatric patients. Can Med Assoc J 1974;110:147–153.
40. Torrey EF. Studies of individuals with schizophrenia never treated with antipsychotic medications: A review. Schizophr Res 2002;58:101–115.
41. McCreadie RG, Padmavati R, Thara R, Srinivasan TN. Spontaneous dyskinesia and parkinsonism in never-medicated, chronically ill patients with schizophrenia: 18-month follow-up. Br J Psychiatry 2002;181:135–137.
42. Owens DG, Johnstone EC, Frith CD. Spontaneous involuntary disorders of movement: Their prevalence, severity, and distribution in chronic schizophrenics with and without treatment with neuroleptics. Arch Gen Psychiatry 1982;39:452–461.
43. Barnes TRE, Rossor M, Trauer T. A comparison of purposeless movements in psychiatric patients treated with antipsychotic drugs, and normal individuals. J Neurol Neurosurg Psychiatry 1983;46:540–546.
44. Delwaide PJ, Desseilles M. Spontaneous buccolinguofacial dyskinesia in the elderly. Acta Neurol Scand 1977;56:256–262.
45. Sulkava R. Alzheimer's disease and senile dementia of Alzheimer type: A comparative study. Acta Neurol Scand 1982;65:636–650.
46. Molsa PK, Marttila RJ, Rinne UK. Extrapyramidal signs in Alzheimer's disease. Neurology 1984;34:1114–1116.

47. Degkwitz R, Wenzel W. Persistent extrapyramidal side effects after long-term application of neuroleptics. In: Brill H, Cole JO, Deniker P, Hippius H, Bradley PB (eds). Neuro-Psycho-Pharmacology. International Congress Series No. 129. Amsterdam: Excerpta Medica Foundation, 1967:608–615.
48. Crane GE. Persistent dyskinesia. Br J Psychiatry 1973;122:395–405.
49. Bourgeois M, Boueilh P, Tignol J. Spontaneous dyskinesia in the elderly and tardive dyskinesia of neuroleptics. A survey among 270 patients [author's transl; in French]. Encephale 1980;6:37–39.
50. Blowers AJ, Borison AL, Blowers CM, Bicknell DJ. Abnormal involuntary movements in the elderly. Br J Psychiatry 1981;139:363–364.
51. Kane JM, Weinhold P, Kinon B, Wegner J, Leader M. Prevalence of abnormal involuntary movements ("spontaneous dyskinesias") in the normal elderly. Psychopharmacology 1982;77:105–108.
52. D'Alessandro R, Benassi G, Cristina E, Gallassi R, Manzaroli D. The prevalence of lingual-facial-buccal dyskinesias in the elderly. Neurology 1986;36:1350–1351.
53. Tublin LM. A treatment for tardive dyskinesia in a complete denture patient. A case report. N Y State Dent J 1989;55:30.
54. Frucht S, Fahn S, Ford B, Gelb M. A geste antagoniste device to treat jaw-closing dystonia. Mov Disord 1999;14:883–886.
55. Kato T, Blanchet PJ, Montplaisir JY, Lavigne GJ. Sleep bruxism and other disorders with orofacial activity during sleep. In: Chokroverty S, Hening WA, Walters AS (eds). Sleep and Movement Disorders. Philadelphia: Butterworth Heinemann, 2003:273–285.
56. Burke RE, Fahn S, Jankovic J, et al. Tardive dystonia: Late-onset and persistent dystonia caused by antipsychotic drugs. Neurology 1982;32:1335–1346.
57. Kiriakakis V, Bhatia KP, Quinn NP, Marsden CD. The natural history of tardive dystonia. A long-term follow-up study of 107 cases. Brain 1998;121:2053–2066.
58. Altrocchi PH, Forno LS. Spontaneous oral-facial dyskinesia: Neuropathology of a case. Neurology 1983;33:802–805.
59. Jankovic J. Tardive syndromes and other drug-induced movement disorders. Clin Neuropharmacol 1995;18:197–214.
60. Sankhla C, Lai EC, Jankovic J. Peripherally induced oromandibular dystonia. J Neurol Neurosurg Psychiatry 1998;65:722–728.
61. Schrag A, Bhatia KP, Quinn NP, Marsden CD. Atypical and typical cranial dystonia following dental procedures. Mov Disord 1999;14:492–496.
62. Tan E-K, Chan L-L, Chang H-M. Severe bruxism following basal ganglia infarcts: Insights into pathophysiology. J Neurol Sci 2004; 217:229–232.
63. Tison F, Louvet-Giendaj C, Henry P, Lagueny A, Gaujard E. Permanent bruxism as a manifestation of the oculo-facial syndrome related to systemic Whipple's disease. Mov Disord 1992;7:82–85.
64. Jankovic J, Beach J. Long-term effects of tetrabenazine in hyperkinetic movement disorders. Neurology 1997;48:358–362.
65. Stip E, Faughnan M, Desjardins I, Labrecque R. Botulinum toxin in a case of severe tardive dyskinesia mixed with dystonia. Br J Psychiatry 1992;161:867–868.
66. Tarsy D, Kaufman D, Sethi KD, Rivner MH, Molho E, Factor S. An open-label study of botulinum toxin A for treatment of tardive dystonia. Clin Neuropharmacol 1997;20:90–93.
67. Kanovsky P, Streitova H, Bares M, Hortova H. Treatment of facial and orolinguomandibular tardive dystonia by botulinum toxin A: Evidence of a long-lasting effect. Mov Disord 1999;14:886–888.
68. Tan EK, Jankovic J. Treating severe bruxism with botulinum toxin. J Am Dent Assoc 2000;131:211–216.
69. Weetman J, Anderson IM, Gregory RP, Gil SS. Bilateral posteroventral pallidotomy for severe antipsychotic induced tardive dyskinesia and dystonia. J Neurol Neurosurg Psychiatry 1997;63: 554–556.
70. Wang Y, Turnbull I, Calne S, Stoessl AJ, Calne DB. Pallidotomy for tardive dyskinesia. Lancet 1997;349:777–778.
71. Trottenberg T, Paul G, Meissner W, Maier-Hauff K, Taschner C, Kupsch A. Pallidal and thalamic neurostimulation in severe tardive dystonia. J Neurol Neurosurg Psychiatry 2001;70:557–559.
72. Sutcher HD, Underwood RB, Beatty RA, Sugar O. Orofacial dyskinesia—A dental dimension. J Am Med Assoc 1971;216:1459–1463.
73. Lauciello F, Appelbaum M. Prosthodontic implications of tardive dyskinesia. N Y State Dent J 1977;43:214–217.
74. Watanabe I, Yamane G, Yamane H, Shimono M. Oral dyskinesia of the aged: II. Electromyographic appearances and dental treatment. Gerodontics 1988;4:310–314.
75. Chiodo GT, Rosenstein DI. Tardive dyskinesia. Gen Dent 1990;38: 289–291.
76. Kai S, Kai H, Tashiro H. Tardive dyskinesia affected by occlusal treatment—A case report. J Craniomandib Pract 1994;12:199–203.
77. Payne AGT, Carr L. Can edentulous patients with orofacial dyskinesia be treated successfully with implants? A case report. J Dent Assoc S Afr 1996;51:67–70.
78. Kelleher MGD, Scott BJJ, Djemal S. Case report: Complications of rehabilitation using osseointegrated implants—Tardive dyskinesia. Eur J Prosthodont Restorative Dent 1998;6:133–136.