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A Rare Case of Spontaneous Peri Oral Dyskinesia: Case Report with Literature Review

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Dyskinesia is an abnormality or impairment of voluntary movement affecting any part of the body. Orofacial dyskinesia involves uncontrolled, stereotypic, paroxysmal movement affecting the face, lips, tongue, jaws or in combination which may lead to severe pain. These involuntary movements are commonly associated with chronic antipsychotic drug usage. But when the dyskinesia occurs with no drug history it is called idiopathic or spontaneous oral dyskinesia. The diagnosis of Spontaneous oral dyskinesia is purely made based on thorough history, precise clinical findings, and necessary investigations to rule out neurological cause. Early management is essential to prevent the Temporomandibular joint and oro-dental complication. Medical management is the first line of treatment in these patients. The following is a clinical diagnostic case report of Spontaneous oral dyskinesia with presenting signs and symptoms, history and examination characteristic of the condition and investigation with successful management.

Keywords: Spontaneous peri oral dyskinesia; tardive dyskinesia; idiopathic; muscle contraction; mandibular disorders.

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ABBREVIATIONS

SOD : Spontaneous Oral Dyskinesia
TD : Tardive Dyskinesia
TMJ : Temporomandibular Mandibular Joint
MRI : Magnetic Resonance Imaging
MRA : Magnetic Resonance Angiography
GABA : Gamma Aminobutyric Acid.

1. INTRODUCTION

There are various movement disorders involving facial, oral or cervical region. One among them is Orofacial dyskinesia. It is an uncoordinated, involuntary, spontaneous, paroxysmal, stereotypic movement of orofacial motor muscular components. It involves the face, lips and tongue and jaws. They often vary in complexity, distribution and severity leading to increased muscle tonicity, inflammation, pain, and fatigue. They may go unnoticed or cause social embarrassment, oral traumatic injuries, speech difficulty, chewing and eating disabilities [1]. Orofacial dyskinesia is of the following types namely, tardive dyskinesia (drug induced), and spontaneous dyskinesia (idiopathic). When there is known history of long-term use of typical antipsychotic drugs that causes various involuntary movements it called Tardive Dyskinesia (TD). The underlying pathophysiology can be dopamine receptor hyperactivity, GABA insufficiency etc. Whereas Oral dyskinesia not associated with use of typical antipsychotic medication its termed as spontaneous oral dyskinesia (SOD) [2].

This is a rare case report of spontaneous perioral dyskinesia which was timely diagnosed and successfully treated with Clonazepam and Trihexyphenidyl hydrochloride.

2. CASE REPORT

A 30 year old female patient, who was a running a small scale business reported to department of oral medicine and radiology post covid pandemic with a chief complaint of spontaneous, uncontrolled, and stereotypic opening of the mouth for past 1 week with pain in the right side of the lower half of the face. Patient gave history of itching over the lower part of face on a fortnight 6 days ago. Following which she cleansed her face with water and cleanser and rubbed vigorously. She immediately developed burning sensation and spontaneous, paroxysmal, involuntary, stereotypic, spasmodic contraction of muscles in lower part of face which led to rhythmic opening of mouth. Patient reported that the paroxysmal movements continued through the day and was absent while sleeping. Following which patient was on carbamazepine 200 mg BD, calcium 500 mg and a drug with combination of mecobalamin, alpha lipoic acid and chromium polynicotinate for 3days and reported there is reduction in frequency of contraction but the symptoms persist. There was no relevant past drug history, medical history or surgical history and family history reported.

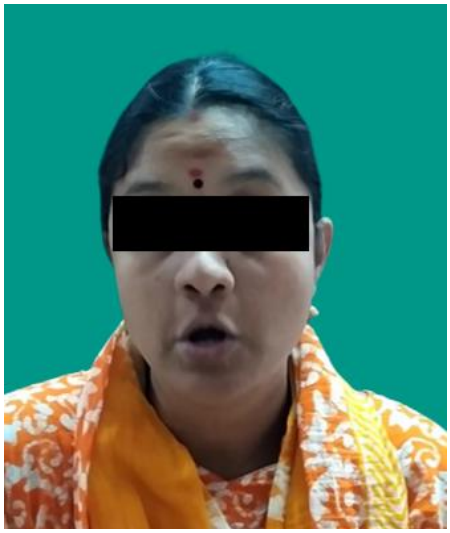

Fig. 1. Extra oral examination

On extraoral examination, inspection there was increase in muscle tonicity in the Orbicularis oris muscle and involuntary downward movement of mandible rhythmically 20 times/ minute which was stereotypic [Fig. 1]. No other dystonic or choreiform movements of extremities present. Patient was apparently healthy with normal mental status. On palpation tenderness was elicited on masseter, lateral pterygoid, medial pterygoid muscles. No sensory loss was observed on face and or oral cavity. On intraoral examination dental caries in 36 and mild gingivitis observed. No evidence of involuntary movement of tongue or hard palate elicited. The patient was subjected to routine blood investigations, which were reported to be normal. MRI and MR Angiography of brain was taken which reported to be normal without any acute hemorrhage or space occupying lesions in brain and neck vessels. So finally correlating history, clinical examination and MRI and MRA a final diagnosis was given as Spontaneous Peri Oral dyskinesia. Further management of the patient was made by prescribing with Clonazepam

0.5 mg BD and Trihexyphenidyl hydrochloride 2 g BD for 1 month. Patient was reviewed after one month and there was complete remission of symptoms and patient was visibly happy without any discomfort.

3. DISCUSSION

Orofacial movement disorders are conditions that affect the motor aspect of the trigeminal, facial, and hypoglossal cranial nerves presenting as hyperactivity or hypoactivity of facial, tongue and masticatory muscles or combinations of these voluntary muscles [3,4]. These movement may also be caused by centrally mediated pathologic conditions involving the basal ganglia and their communication with other areas of the brain which is responsible for fine motor functions. Orofacial movement disorders include tardive dyskinesia, spontaneous oral dyskinesia (SOD), sleep bruxism, oromandibular dystonia, Parkinson disease, psychogenic movement disorder. The term orofacial dyskinesia involves involuntary, repetitive, stereotypical movement of face, lip, tongue, jaws that may be painful. It may also be associated involuntary eyelids movements thereby causing blinking or sustained closure of eyelids during involuntary mandibular movement.

When patient gives history of chronic usage of typical antipsychotic drug therapy which are potent dopamine D2 receptor blocker it is called Tardive dyskinesia which is average estimated to be 20% cases. Length of antipsychotic drug exposure of at least 3 months in younger individuals and short of 1 month in elderly individuals is reported [1]. Orofacial dyskinesia not associated with use of any antipsychotic drugs were recognized and termed as spontaneous orofacial dyskinesia (SOD) by Varga et al. in 1982 [2]. The clinical presentation of SOD involving various combinations of tongue, lips and jaw movements are milder in intensity when compared with tardive dyskinesia [4]. SOD was reported by Degkwitz and Wenzel in 1 % of non demented elderly subjects and demented institutionalized patients [5]. A prevalence rate of 12% is seen in individuals with chronic schizophrenia who have been never treated [6]. A study by Owen et al was on oral dyskinesia patients with the schizophrenia who had with and without drug exposure and observed severe oral dyskinesia in patients with drug usage [7]. Presence of SOD observed in other neurological disorders such as autism, mental retardation, Alzheimer's disease and Rett syndrome seems to be non-specific [8]. Edentulous dyskinesia was earlier considered as one subtype of SOD seen in patients with no replacement of tooth after extraction and ill-fitting dentures. Stereotyped smacking, pursing of lips, lateral deviation and protruding jaw movements documented in these patients. In such cases, oro dental factor may be the trigger for oral dyskinesia. They can be managed by prosthetic correction of the dental abnormalities. P.J Blanchet et al. studied prevalence of SOD in elderly and concluded it is more common in elderly than Tardive dyskinesia [9-13].

In very rare conditions of 1-2% dyskinesia occurs in the absence of drug exposure and diseases known to be associated. Women are commonly affected than men and young age group is very rarely affected [14]. In our case the patient was a young woman, who was in apparently healthy and good mental status, without any previous antipsychotic drug exposure or any predisposing diseases which is very rare condition. Only muscles of peri oral structure were affected without tongue or facial involvement. So, we consider it the very rarest case of spontaneous perioral dyskinesia.

Proper and early management is essential in patients with oral dyskinesia or oromandibular movement disorders because it may lead to complications such as TMJ degenerative changes, muscle stiffness, mucosal lesions, damage to teeth and dental prosthesis which eventually affect the quality of life of patients [15]. Medical management is done with Anticholinergics (Trihexyphenidyl hydrochloride), GABAergic (Baclofen), Benzodiazepines (Clonazepam, Diazepam), Dopaminergic (Levodopa), Antiparkinsonians (Amantadine, Diphenhydramine, Benztropine)and Non benzodiazepines (Buspirone) [4,16]. Chemo-denervation with botulinum neurotoxin and surgery other modalities of treatment when medical management does not seem to be effective. In our case successful management was done with Clonazepam and trihexyphenidyl hydrochloride. Clonazepam is a long-acting benzodiazepine which acts by modulation of GABA receptor, thereby enhancing GABAergic inhibition of neuronal firing. Fukasawa et al. [2] in 2001 reported that clonazepam can be successfully used in patients with spontaneous oral dyskinesia without tolerance. Trihexyphenidyl is antiparkinsonian used to decrease muscle stiffness/ rigidity. It is

anticholinergic acts by blocking acetylcholine in the nerve terminal.

4. CONCLUSION

A complete oral health is not only the field of interest for a dentist but also dentist must be familiar with orofacial dyskinesia to successfully manage the condition of these patients who are clueless of sudden involuntary movement of the stomatognathic system. Proper and early management of orofacial dyskinesia can prevent further orodental and TMJ complications. Spontaneous oral dyskinesia can be diagnosed if there is no drug history or any associated diseases. From our case we are reporting that orofacial dyskinesia not only occurs in elderly but also young healthy individuals. Early diagnosis in our case was the key of successful management and preventing further complications and improving quality of life of patient.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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