SPONTANEOUS PERI ORAL DYSKINESIA- A RARE CASE REPORT

ABSTRACT:

Dyskinesia is an abnormality or impairment of voluntary movement affecting any part of body. Orofacial dyskinesia involves uncontrolled, stereotypic, paroxysmal movement affecting face, lips, tongue, jaws or in combination which may lead to severe pain. They are commonly associated with chronic antipsychotic drug usage but when no history is relevant to cause the sudden involuntary movement spontaneous oral dyskinesia must be considered. The diagnosis of Spontaneous oral dyskinesia is purely made based on clinical and and thorough proper history taking and investigations to rule out neurological abnormality. Early management is essential to prevent TMJ and orodental 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.

KEY WORDS: spontaneous perioral dyskinesia, idiopathic dyskinesia, involuntary, muscle contraction, mandibular movement disorders.

ABBREVIATION: SOD: spontaneous oral dyskinesia, TMJ: Temporomandibular mandibular joint, MRI: Magnetic resonance imaging, MRA: Magnetic resonance angiography, GABA: Gamma Aminobutyric Acid.

INTRODUCTION:

Orofacial dyskinesia is an uncoordinated, involuntary, spontaneous, paroxysmal, stereotypic movement of orofacial motor muscular components. It involves 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. Orofacial dyskinesia is of following types namely, Tardive dyskinesia (drug induced), Spontaneous dyskinesia (idiopathic).

CASE REPORT:

A 30year old female patient, who was a running a small lscale 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 200mg BD, calcium 500mg 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.

FIGURE 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. [Figure 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. Patient was subjected to routine blood investigation done, which ported 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 0.5mg BD and Trihexyphenidyl hydrochloride 2g 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.

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. [8,15] 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.[5] The clinical presentation of SOD involving various combinations of tongue, lips and jaw movements are milder in intensity when compared with tardive dyskinesia. [15] SOD was reported by Degkwitz and Wenzel in 1 % of non demented elderly subjects

and demented institutionalized patients.[3] 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. [13] Presence of SOD observed in other neurological disorders such as autism, mental retardation, Alzheimer's disease and Rett syndrome seems to be non-specific. [14] 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. [4] [9-12]

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.[2] 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.[7] Medical management is done with Anticholinergics (Trihexyphenidyl hydrochloride), GABAergic (Baclofen), Benzodiazepines (Clonazepam, Diazepam), Dopaminergic (Levodopa), Antiparkinsonians (Amantadine, Diphenhydramine, Benztropine)and Non benzodiazepines (Buspirone). [15,16] 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. Fukesama et al in 2001 reported that clonazepam can be successfully used in patients with spontaneous oral dyskinesia without tolerance.[5] Trihexyphenidyl is antiparkinsonian used to decrease muscle stiffness/ rigidity. It is anticholinergic acts by blocking acetylcholine in the nerve terminal.

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

PATIENT CONSENT
Obtained

ETHICAL APPROVAL
It is not applicable.

COMPETING INTERESTS
Authors have declared that no competing interests exist

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