

## CASE REPORT

# Oro-Mandibular Dystonia Developing with both Sertraline and Fluoxetine Use in a Female Adolescent with Major Depressive Disorder

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## Abstract

Oro-mandibular dystonia (OMD) involves repetitive uncontrollable contractions of the lower facial muscles (Scorr et al. 2021). Its etiology is not fully elucidated although various neurological (encephalitis, etc.), psychogenic and psychopharmacological (SSRIs, antipsychotics etc.) causes were implicated (Kutuk et al. 2019, Scorr et al. 2021). The odds ratio (OR) is 1.7 for dystonia in general after using SSRIs (Revet et al. 2020). Paroxetine (OR= 1.7) and fluoxetine (OR= 1.5) have the highest risks among SSRIs while escitalopram has the lowest (OR=0.8, Revet et al. 2020). Adolescent patients developing OMD after treatment with sertraline or fluoxetine alone were reported previously (Kutuk et al. 2019, Revet et al. 2020). However, OMD in a patient with sequential trials of different SSRIs are not reported up to now.

**Keywords:** Oro-mandibular Dystonia, Fluoxetine, Sertraline, Major Depressive Disorder

## CASE

A 13-years old female was referred to the clinic for “sadness and depression”. The symptoms started seven months ago after inter-parental conflict and included anhedonia, social withdrawal, and feelings of guilt. Her medical history was normal while her mother was treated for major depressive disorder (MDD) three years ago. Mental status examination revealed depression. Children’s Depression Inventory (CDI) score was 20 (above the cut-off). She was diagnosed with MDD according to DSM-5 criteria (Clinical Global Impressions Scale – Severity, CGI-S=4, moderate). Cognitive Behavioral and Family Therapies (CBT/ FT) were planned while sertraline was initiated at 25 mg/ day. The dose was increased to 50 mg/ day in the second week. She was brought to the clinic due to involuntary contractions

of the lower face which started in the second month. The Abnormal Involuntary Movements Scale (AIMS) score was 15 while neurological examination demonstrated painful mastication and lateralization of the tongue.

Other neurological/ physical examinations, laboratory investigations, an Electroencephalogram (EEG), and cranial Magnetic Resonance Imaging (MRI) were normal. Sertraline was suspected in etiology and the dose was reduced to 25 mg/ day. After the fourth day her complaints were reduced (AIMS= 5). She complained of limited efficacy after a month at this dose (CDI= 27, CGI-S= 4) and was started on fluoxetine 20 mg/ day. The dose was titrated to 40 mg/ day in the second month and OMD re-emerged a week later (AIMS= 10). The complaints were remitted again a week later with a reduced dose (i.e. 20 mg/ day). Evaluation with the Naranjo algorithm (Naranjo et al. 1981) revealed a score of 8 for both agents (probable). In the follow-up, the patient’s treatment was continued with fluoxetine with a dose of 20mg/day, and Cognitive Behavioral Therapy (CBT). Her complaints about the involuntary contractions of her lower face haven’t recurred since then. Her Informed consent was obtained from the legal guardian of the patient for publication.

Like most previous reports of OMD with SSRIs, our patient was female, and the complaints were reversible/

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dose-related and emerged sometime after treatment (Kutuk et al. 2019, Revet et al. 2020, Scorr et al. 2021). Family history was negative and no stressors and/ or neurological/ medical conditions could be found. There were no additional treatments and relationships with both agents were judged “probable”. The mechanism of SSRIs causing dystonia is not fully understood. The most supported hypothesis is that OMD after sertraline/ fluoxetine may emerge due to increased serotonergic neurotransmission in basal ganglia and a relative dopaminergic deficit or may involve other mechanisms (Kutuk et al. 2019). Serotonin transporter inhibition can alter the regulation of the enzyme which is playing role in dopamine biosynthesis in dopaminergic neurons in the substantia nigra and these changes may play a role in dystonia (MacGillivray et al. 2011). Also, the cerebellum could have a possible role in the mechanism of dystonia (Gonzalez et al. 2014). Another hypothesis asserts that some changes in the balance between serotonergic, dopaminergic, cholinergic, and noradrenergic tones may have a role in the mechanism of SSRI-induced dystonia (Madhusoodanan et al. 2010). Despite its rarity, this adverse effect may be borne in mind by clinicians.

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