

Case Report**Oropharyngeal dysphagia in extrapyramidal syndrome****Ferdinand Simanjuntak*, Rery Budiarti**, Muyassaroh*****Department of Oto Rhino Laryngology-Head and Neck Surgery,
Faculty of Medicine Universitas Diponegoro**Regional General Hospital Dr.Kariadi,
Semarang**ABSTRACT**

Background: Dysphagia could be caused by mechanical or motoric problems. Dysphagia is associated with neurological disorders in extrapyramidal syndrome (EPS), including tardive dyskinesia caused by long-term chronic use of antipsychotic drugs. Dysphagia in EPS about 9.4% occurs in young adults. Dysphagia in EPS could be a single symptom or as an concomitant symptom. **Purpose:** To discuss a case of oropharyngeal dysphagia in EPS due to the effects of antipsychotic drugs on a patient who was treated under multidisciplinary management. **Case report:** A 32-year-old male patient came to Emergency Unit of Dr.Kariadi Hospital with chief complaint of a sudden onset of difficulty in swallowing liquid and solid food. He had a history of longterm use of Chlorpromazine. The diagnosis of oropharyngeal dysphagia was established with Fiberoptic Endoscopic Evaluation of Swallowing (FEES). **Clinical question:** What is the proper management for dysphagia in EPS? **Method:** Literature search through database Pubmed and Google Scholar found 88 journals. **Results:** There were 4 journals relevant with the case. Those journals reviewed the diagnosis and disclosed the management of EPS with dysphagia was discontinuation of the analogous antipsychotic drugs and swallowing practice physiotherapy. **Conclusion:** Dysphagia in this case was an EPS due to chlorpromazine use, and made a good respond after drug replacement.

Keywords: oropharyngeal dysphagia, extrapyramidal syndrome, chlorpromazine**ABSTRAK**

Latar belakang: Disfagia dapat disebabkan oleh gangguan mekanik maupun motorik. Disfagia dapat berhubungan dengan kelainan neurologis pada sindrom ekstrapiramidal (SEP), antara lain tardive diskinesia yang disebabkan pemakaian obat antipsikotik jangka panjang. Disfagia pada SEP sekitar 9,4 % terjadi pada usia dewasa muda. Disfagia pada SEP dapat merupakan gejala tunggal atau sebagai gejala penyerta. **Tujuan:** Membahas kasus disfagia orofaringeal pada SEP akibat efek obat antipsikotik pada pasien yang diterapi secara multidisipliner. **Laporan kasus:** Seorang laki-laki usia 32 tahun datang ke IGD RSUP Dr. Kariadi dengan keluhan utama mendadak tidak dapat menelan sama sekali, baik cairan maupun makanan padat. Didapatkan tanda gejala SEP dengan riwayat pemakaian chlorpromazine jangka panjang. Diagnosis disfagia orofaringeal ditegakkan dengan pemeriksaan Fiberoptic Endoscopic Evaluation of Swallowing (FEES). **Pertanyaan klinis:** Bagaimana penatalaksanaan yang tepat untuk kasus disfagia pada sindrom ekstrapiramidal? **Metode:** Penelusuran kepustakaan melalui database Pubmed dan Google Scholar diperoleh 88 jurnal. **Hasil:** Didapatkan 4 jurnal yang relevan dengan kasus. Empat jurnal tersebut menyatakan tatalaksana disfagia dengan SEP ialah dilakukan penghentian obat antipsikotik tipikal dan latihan menelan. **Kesimpulan:** Disfagia pada SEP akibat penggunaan chlorpromazine memberikan respon baik setelah dilakukan penggantian obat.

Kata kunci: disfagia orofaringeal, sindrome ekstrapiramidal, chlorpromazine

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INTRODUCTION

Oropharyngeal functional dysphagia could emerge as the result of an abnormality in the oral or pharyngeal cavity. Dysphagia may cause tracheobronchial aspiration, malnutrition, or dehydration. Up to 43%-50% dysphagia presents aspiration pneumonia with 45% mortality rate. It is estimated that 30%-50% got aspiration pneumonia in the acute phase, 5% in the chronic state, and 38-48% had silent aspiration after 2-3 months of having dysphagia. The highest prevalence is in 5-6th decades, and very rare in a young person (15-35 years of age), reported in only about 9.4%. Oropharyngeal dysphagia could be caused by problems of the teeth or oral mucosa, mechanical obstruction, neuromuscular, or longterm use of antipsychotic drugs.^{1,2}

Antipsychotic drugs could generate extrapyramidal syndrome which appear as acute involuntary movement (Parkinson, acute dystonia, acute ataxia) or chronic tardive dyskinesia (tardive syndrome). Tardive dyskinesia (TD) is a serious, often disabling movement disorder caused by medication that blocked dopamine receptors. Extrapyramidal syndrome more often happens in patients consuming the typical or first-generation anti psychotics (FGA) drugs, and less common in second generation anti psychotics (SGA).³ Dysphagia in extrapyramidal syndrome cases need to be evaluated thoroughly and provided multidisciplinary management. Evaluation of dysphagia should include swallowing function, the affected anatomical level, and the underlying disease. The management of extrapyramidal syndrome includes multimodality and multidisciplinary collaboration.^{4,5}

The purpose of this presentation was to discuss oropharyngeal dysphagia in extrapyramidal syndrome as the effect of antipsychotic drugs on a patient who had been treated under multidisciplinary management

CASE REPORT

A 32 years old male came to The Emergency Unit of Dr Kariadi General Hospital with chief complaint difficulty of swallowing. All of a sudden he was totally unable to swallow, solid food and also liquid. He felt something stuck in his throat. He often had throbbing headache at the back part of his head, which worsened with physical activity, and decreased when he rest. He also had an additional symptom of tremor in his right hand, especially during strenuous physical activity, and emotional strain. He often went to district health center and was prescribed tranquilizer. There was no history of nausea, vomitus, seizure, and loss of consciousness. There was no fever, and coughing.

In the last 2 weeks, he often experienced difficulty in swallowing, harder for solid food such as steamed rice, but easier for porridge. He had been taking tranquilizer chlorpromazine prescribed by the doctor in district health center for 3 months.

Physical examination revealed a good general condition, compos mentis, blood pressure 140/90 mmHg, heart rate 80x/minute, respiration rate 16x/minute, temperature 36.8°C, heart and lung auscultation within normal limits. Throat examination disclosed symmetric pharyngeal arches, uvula position in the middle of the soft palate, no tongue deviation, and there was a decreased of gagging reflex. The ear and nose were within normal limits.

Electromyography examination of nerve conduction velocity (NCV) affirmed motoric mononeuropathy of the left ulnar nerve. Sensory conduction velocity (SCV) within normal limits, repetitive nerve stimulation (RNS) showed sign of myasthenia gravis, which need clinical confirmation. The result of examination for spasmophilia was strong positive, Chvostek sign positive, carpal tunnel syndrome positive. Thorax photo showed normal heart and lung.

FEES (*fibre optic endoscopic evaluation of swallowing*) pre swallowing assessment conducted on February 19, 2020 revealed: right side leakage on inflation of the cheeks, asymmetric uvula movement drawn to the left, coughing strength within normal limit. There was right side leakage on velopharyngeal movement, and decreased muscle tone of the right lateral pharyngeal wall. On swallowing assessment, there were residual foodstuff in all consistencies, no sign of silent aspiration in all bolus consistencies, and coughing reflex positive.



Figure 1. Thorax X-ray. No lung infiltrate and nodule. No cardiomegaly



Figure 1. Thorax MSCT with contrast. No mass in the lung and mediastinum.

Thorax multi-slice computed tomography (MSCT) with contrast was performed on February 21, 2020 to exclude thymoma. The result showed no mass in the mediastinum and the lung.

The conclusion was oropharyngeal phase dysphagia, caused by neuromuscular abnormality. The differential diagnosis was extrapyramidal syndrome and myasthenia gravis. It was presumed that the swallowing problem and tremor of the right hand was the result of long term administration of antipsychotic medication.

The management was discontinuation of chlorpromazine as typical antipsychotic drug, replaced by clozapine (atypical antipsychotic drug) by the neurologist. It was followed by swallowing exercise under medical rehabilitation specialist, and consultation to a psychiatrist.

CLINICAL QUESTION

What is the proper management for dysphagia in EPS?

METHOD

Literature search in 2015 and 2020 through database Pubmed and Google Scholar using keyword “oro-pharyngeal dysphagia” AND “extra pyramidal syndrome” AND “chlorpromazine”, obtained 88 journals. Out of 88 literatures, 4 journals were relevant with the inclusion criteria. The inclusion criteria were: oropharyngeal dysphagia, extrapyramidal syndrome, tardive dyskinesia, chlorpromazine. The exclusion criteria were: stroke, oesophageal dysphagia, peripheral neuropathy.

RESULT

There were 4 journals relevant with the case. Those journals reviewed the diagnosis,

and disclosed that the management of EPS with dysphagia was discontinuation

of the analogous antipsychotic drugs and swallowing practice physiotherapy.

Table 1. Literature search in case reports

No.	Researcher-journal	Patient characteristics	Supporting examination	Measure	Evaluation post-measure
1.	Kadota Y, Hori H, Takayama M, Okabe C, Ohara N. ⁴	Female, 61 y.o tardive dyskinesia with schizophrenia.	Oropharyngeal dysphagia after antipsychotic use.	Stop typical antipsychotic drugs.	Dystonia ceased 3 days after drug discontinuation.
2.	Gregory RP, Smith PT, Rudge P. ⁵	Female, 48 y.o, dysphagia for 6 years. History of flupenthixol and chlorpromazine	Video-fluoroscopic barium swallow	Stop antipsychotic drugs for 10 months	Barium swallow video-fluoroscopic, showed increased ability of swallowing solidified food.
3	Bhat PS, Pardal PK, Diwakar M. ⁶	Male, 37 y.o, schizophrenia. Typical antipsychotic drug injection. Inconsistent administration.	Normal oropharynx, larynx and oesophagus	Replaced medication with atypical antipsychotic drugs.	Dysphagia resolved.

Table 2. Literature review of research study

No.	Researcher	Number of patients	Diagnosis	Measure	Evaluation post measure
1.	Miarons M, Rofes L. ⁷	14 patients	Dysphagia in typical antipsychotic drugs	Change to other antipsychotic drugs.	Improved symptoms
		22 patients	Dysphagia in atypical antipsychotic drugs	Change to other antipsychotic drugs.	Improved symptoms

DISCUSSION

Oropharyngeal dysphagia in adults could be generated by neurological dysfunction such as in stroke cases, peripheral neuropathy in larynx, pharynx, tongue and jaw regions, myasthenia gravis, or myopathy. It could also caused by head and neck structural changes in cancer, post-surgery cases, chemo or radiotherapy, iatrogenic trauma, or congenital abnormality, and other causes such as infection, laryngopharyngeal reflux, systemic disease, side effect of drugs, and psychogenic.⁸

Swallowing process is a complex neuromuscular activity which involved a swift

coordination of structures in the oral cavity, pharynx, larynx and oesophagus. During swallowing process, bolus of food or fluid will be moved from the mouth into the gaster through the pharynx and oesophageal tract. The whole process is involving about 40 pairs of muscle and 5 cranial nerves.^{1,2,8}

Patients with dysphagia symptom need a comprehensive management, starting with history taking, physical examination including neurological examination and supporting investigation. Laboratory test such as *acetylcholine receptor* (AChR) detection can help determined the causal of dysphagia in myasthenia gravis (MG); measurement of serum muscle enzyme for inflammatory my-

opathy, or thyroid function test to establish the diagnosis of toxic myopathy. Supporting examination including *videofluoroscopic swallowing study* (VFSS), *fiberoptic endoscopic evaluation of swallowing* (FEES), *upper endoscopy (esophagogastroduodenoscopy)*, and oesophageal manometry.^{9,10}

Extrapyramidal syndrome is symptoms of the extrapyramidal nerve tract disorder which emerge as acute involuntary movement (Parkinson, acute dystonia, acute ataxia) or chronic tardive dyskinesia (tardive syndrome). Tardive dyskinesia (TD) is a serious, often disabling movement disorder, such as stereotype/classical oro-buccal-lingual-facial dyskinesia, tardive akathisia, tardive dystonia, tardive chorea, tardive tremor and tardive Parkinsonism, tardive myoclonus, tardive oculogyric deviations, tardive tics, tardive Gilles de la Tourette's syndrome, tardive pain, withdrawal emergent syndrome. Extrapyramidal syndrome more often happens in patients consuming the typical or first-generation anti psychotics (FGA) drugs, and less common in second generation anti psychotics (SGA). Even though tardive dyskinesia (TD) or tardive syndrome cases are rare, the management or prevention is more challenging.^{3,4,11,12}

In our patient, there was an involuntary movement and abnormal motoric system reflex, drooping eyelids, tremor of the hands, and difficulty in swallowing.

Tardive dyskinesia or tardive syndrome is a medication-induced movement disorder as an effect of long term or high dose neuroleptic drugs administration. The symptoms include akathisia, dystonia, oro-buccal-lingual-facial (OBLF): chewing movements, lips sucking, lips smacking, tongue protrusion, fast eyes blinking, facial grimacing. Other symptoms such as limb-truncal syndrome: choreiform/choreoathetoid movements of the fingers, hands, upper or lower extremities, and sometimes a stretching motion of the trunks or mix movements. The symptoms usually

appear after 3 months using neuroleptic or antipsychotic drug, and in some rare cases tardive dyskinesia come up after 4 weeks consuming oral antipsychotic drug, or after 8 weeks antipsychotic injection administration.¹²⁻¹⁵

Neuroleptic drugs which frequently generate tardive dyskinesia are typical neuroleptic type or first-generation antipsychotics (FGA) such as haloperidol, perphenazine, trifluoperazine, fluphenazine, and less often chlorpromazine, thioridazine, pimozide. Comparatively the atypical or second -generation antipsychotics (SGA) or also known as serotonin 5HT2 and dopamine D2 receptor antagonist was less frequent causing TD such as olanzapine, risperidone, aripiprazole, quetiapine, and clozapine was extremely rare and could diminish the tardive dyskinesia risk compared to the other drugs.^{3,6,8,12}

This patient had a history dysp of dysphagia after consuming psychotropic drugs more than 4 weeks, caused by mental stress and felt better after taking the drugs. Dysphagia due to taking antipsychotic drugs had been reported by Kadota et al.⁴, Gregory et al.⁵, Miaron et al.⁶, and Bhat et al.⁷ The symptom of dysphagia clear up after discontinuation of the drugs.

Our case was a 32 years old male with oropharyngeal dysphagia due to extrapyramidal syndrome as the side effect of medication with chlorpromazine, a FGA drug. The management of the case was discontinuation of chlorpromazine and replaced with SGA drugs aripiprazole, quetiapine, and clozapine, along with swallowing practice physiotherapy. The multidisciplinary treatment brought a good result, the patient could swallow liquid and solid food normally.

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