DOI:10.14744/bej.2022.93446
Beyoglu Eye J 2022; 7(3): 237-239

Case Report

Temporomandibular Joint Dislocation in a Patient with Keratoconus Under General Anesthesia: A Case Report

Mukaddes Damla Ciftci,[1] Tuba Kuvvet Yoldas,[2] Canan Bor,[2] Melis Palamar[1]

[1]Department of Ophthalmology, Ege University, Izmir, Türkiye
[2]Department of Anesthesiology and Reanimation, Ege University, Izmir, Türkiye

Abstract

Keratoconus is a common disease in which the central or paracentral cornea undergoes progressive thinning, resulting in a cone-shaped cornea. It has been associated with many systemic disorders such as Down syndrome, osteogenesis imperfecta, and joint hypermobility. Herein, temporomandibular joint dislocation under general anesthesia during supraglottic airway device placement before deep anterior lamellar keratoplasty procedure in a keratoconus patient is reported.

Keywords: Anesthesia, cornea, joint hypermobility, keratoconus, keratoplasty

Introduction

Keratoconus is a common disorder characterized by progressive corneal steepening with eventual corneal thinning. Its prevalence is 50–230 cases/100,000 (1). It has been associated with many systemic disorders such as atopic disease, Down syndrome, osteogenesis imperfecta, joint hypermobility, and mitral valve prolapse (2). Although it is known that these patients have hyperelastic joints, temporomandibular joint (TMJ) dislocation is not a common finding. Predisposing factors for TMJ dislocation include congenital joint weakness, extreme mouth opening during yawning, dental and otorhinolaryngological treatment, trauma, and drugs, especially the anti-emetics (metoclopramide) and phenothiazines (Compazine), which produce extrapyramidal effects, hypermobility, associated with systemic diseases, and psychogenic and neurological disorders (3).

Herein, bilateral TMJ dislocation in a keratoconus patient under general anesthesia during supraglottic airway device placement before deep anterior lamellar keratoplasty (DALK) is reported.

Case Report

A 25-year-old Caucasian female with keratoconus was referred to our clinic. Her best-corrected visual acuity (BCVA) in both eyes was 1.8 log MAR. Slit-lamp anterior segment examination revealed a hydrops scarring in the left cornea (Fig. 1). The patient had no family history of keratoconus. There was no history of allergic conjunctivitis and contact lens use. BCVA tried with contact lenses was 1.8 log MAR. She was using glasses for irregular astigmatism before surgery.

She was electively scheduled for DALK under general anesthesia. Routine pre-operative investigations, patient medical history, physical examination, and laboratory results were un-

How to cite this article: Ciftci MD, Kuvvet Yoldas T, Bor C, Palamar M. Temporomandibular Joint Dislocation in a Patient with Keratoconus Under General Anesthesia: A Case Report. Beyoglu Eye J 2022; 7(3): 237-239.

Address for correspondence: Melis Palamar, MD. Department of Ophthalmology, Ege University, Izmir, Türkiye
Phone: +90 530 408 50 51 **E-mail:** melispalamar@gmail.com

Submitted Date: February 28, 2022 **Accepted Date:** May 21, 2022 **Available Online Date:** August 05, 2022

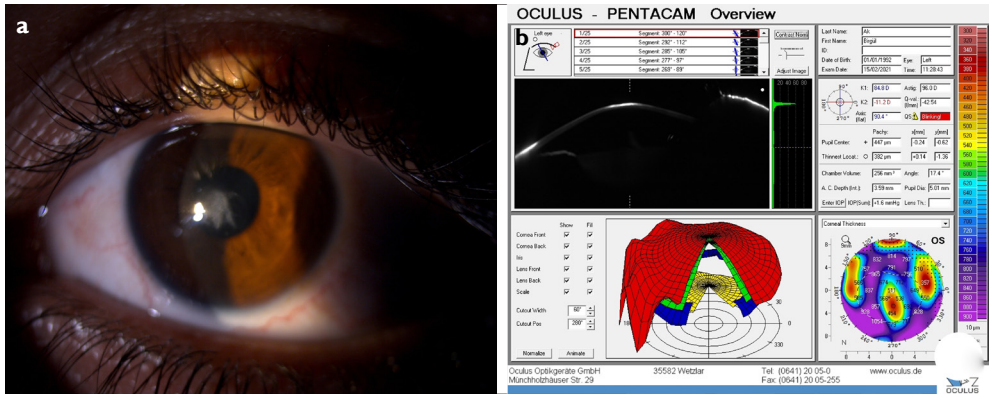

Figure 1. (a) Pre-operative anterior segment photograph demonstrates hydrops scarring. **(b)** Corneal topography shows irregular astigmatism.

remarkable. Airway management was evaluated, the mouth opening, neck movements were adequate, and airway was graded as Mallampati Class II. In the operation theater, after standard monitors pulse oximeter (SpO_2), ECG, NIBP, and $EtCO_2$ were attached, anesthesia was induced with intravenous 0.01 mg/kg atropine (Atropine Sulfate, 0.5 mg, Galen, Türkiye), 2 mg/kg propofol (Propofol PF 1% 200 mg/20 ml, Polifarma, Türkiye), 0.6 mg/kg rocuronium (Esmeron 50 mg/5 ml, MSD, Netherlands), and 1 mcg/kg fentanyl (Talinat 0.5 mg/10 ml, Vem, Türkiye). The patient was pre-oxygenated with 80% oxygen + 20% air. After achieving adequate muscle relaxation, lubricated second-generation supraglottic airway device (I-gel, Intersurgical, Lithuania, USA, size: 4, with a non-inflatable cuff and hole a gastric catheter features) was inserted by assisting anesthesiologist, grasping the jaw, and lifting it upward (jaw-thrust maneuver – a method for opening the airway). Supraglottic airway device was preferred because of the short operation time. Furthermore, endotracheal intubation and extubation are associated with coughing, increased heart rate, elevated blood pressure, and, eventually, raised intraocular pressure which is undesirable in ophthalmic surgical patients (4). While inserting I-gel, the anesthesiologist noticed that the patient's jaw had been deformed and protruded anteroinferiorly. Patient's jaw was locked wide open in a protracted position and bilateral anterior TMJ dislocation was diagnosed. After insertion of I-gel and successful ventilation, anesthesia was maintained with 2% sevoflurane (Sevorane Liquid 100%, Abbvie, England) in a mixture of O_2 and air in 40:60 ratios to maintain a minimum alveolar concentration of 1. After a successful DALK procedure, while the patient was still under anesthesia, the TMJ dislocation was reduced manually by applying an inferior and posterior force to slide the condyle back into the glenoid fossa. Two clicks were felt as dislocation was corrected. After correction of TMJ dislocation, intravenous sugammadex (Bridion 200 mg/2 ml, MSD, USA) 2 mg/kg was administered to gain spontaneous ventilation. The total surgical procedure was completed in 50 min. On awakening, the patient was able to close her mouth, move her jaw without difficulty, and talk normally. Paracetamol (Paracerol, 10 mg/ml, Polifarma, Türkiye) 10 mg/kg was given for both post-operative analgesia and joint pain. After recovery in post-anesthesia care unit, the patient has been questioned for TMJ dislocation history. There was no history of dislocation, trauma to face, and any connective tissue disorder or neuromuscular disorder. The patient was evaluated using Beighton score (5) that examining flexibility of some joints and giving a maximum of 9 points. The patient's Beighton score was 4 points. She had benign joint hypermobility syndrome according to this score. All procedures of anesthesia were performed by an expert anesthesiologist who has 14 years of experience and has never encountered such a situation before. When all general anesthesia cases were evaluated in our center, there were only three cases of TMJ dislocation in the past 20 years.

The patient was informed about this publication and written consent was obtained for publishing medical records.

Discussion

Keratoconus may not be a distinct ophthalmologic disease, but a sign representing a systemic disorder, possibly a mild collagen tissue abnormality. The patient experienced TMJ dislocation due to a possible collagen tissue abnormality even if she did not have any other predisposing factor. Because the patient did not have dermatological, auditory, and other systemic findings that suggest Ehlers-Danlos syndrome or Brittle Cornea syndrome, genetic analysis did not considered.

The present patient was examined for existence of hypermobility using Beighton score (5) and Helkimo Clinical Dysfunction Index (HCDI) (6). Beighton score is used to determine generalized joint hypermobility. HCDI is a simple and quick test used to evaluate subjects affected by temporomandibular disorders. The test evaluates movement, joint function, pain, and musculature, providing a quick general overview that could be very useful at different levels of care. Beighton score was 4 points and pointed out generalized joint hypermobility.

Generalized joint hypermobility is known to increase the risk of TMJ disorders (7). HCDI was 4 points and suggested mild TMJ dysfunction. TMJ dislocation may not be anticipated in a patient without a history of TMJ instability and hypermobility existence. These tests are not routinely performed, and they were performed in the present patient to evaluate TMJ disease in the post-operative period.

Joint hypermobility is not a diagnosis, but is a term used to define a joint that exceeds its normal range of motion, taking into account age, sex, and race. African, Asian, and Middle Eastern individuals are known to have increased joint laxity. This feature is predominantly determined by the tightness or laxity of ligaments, which, in turn, is influenced by genetics, involving the connective tissue genes collagen, elastin, and fibrillin. Benign joint hypermobility syndrome is a connective tissue disorder with hypermobility in which musculoskeletal symptoms occur in the absence of systemic rheumatological diseases. Heritable connective tissue disorders such as Ehlers-Danlos syndrome, Marfan syndrome, and osteogenesis imperfecta result in systemic ligamentous laxity and cause local joint hypermobility and generalized joint laxity. The Beighton score is used as the standard method for the hypermobility syndromes. Our patient has benign joint hypermobility according to Beighton score (5).

TMJ dislocation is a rare but reported complication of supraglottic airway device use. Sia et al. (8) reported two cases in whom TMJ dislocation after laryngeal mask airway insertion. They highlighted three factors that predispose to anterior dislocation: Poor joint capsule integrity, poor joint superiority morphology, and muscle hypotonicity. Acute anterior dislocation of the TMJ might be detected as a result of laughing, taking a big bite, trauma, convulsions, yawning, and induction of anesthesia. Muscle relaxation leading to hypotonicity of jaw muscles and jaw trust maneuver may result in TMJ dislocation. Possible precipitating factors include the effect of the muscle relaxant, direct laryngoscopy, and placement of an oral airway; jaw-thrust maneuver, passive wide mouth opening, and loss of muscle tone, have all been described as factors that could lead to TMJ displacement under anesthesia. In the present case, joint hypermobility and general anesthesia both caused TMJ dislocation. One of the limitations of this study is not being able to rule out that general anesthesia may cause TMJ dislocation. Despite all this, it cannot be excluded that TMJ joint dislocation and keratoconus may coincidentally exist in the same patient.

Street et al. (9) found that there was no statistically significant difference in the prevalence of hypermobile joints in keratoconus patients and controls. In contrast, Woodward and Morris. (10) found that patients with keratoconus are 5 times more likely to show hypermobility of the metacarpophalangeal and wrist joints. Galperin et al. (11) reported a case of keratectasia after laser in situ keratomileusis in both eyes in a patient with benign joint hypermobility syndrome.

As a conclusion, patients with keratoconus might have a tendency to have hypermobile joints. These hypermobile joints may lead to surprising joint dislocations during general anesthesia. Both anesthesiologists and ophthalmologists should be aware of a possible TMJ dislocation in keratoconus cases and also ensure early management.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

Authorship Contributions: Concept – M.P.; Design – M.P., C.B.; Supervision – M.P., C.B.; Materials – T.K.Y.; Data collection and/or processing – M.D.Ç.; Analysis and/or interpretation – M.D.Ç., T.K.Y.; Literature search – M.D.Ç., T.K.Y.; Writing – M.D.Ç., T.K.Y.; Critical review – M.P., C.B.

References

1. Bechrakis N, Blom ML, Stark WJ, Green WR. Recurrent keratoconus. Cornea 1994;13:73–7. [CrossRef]
2. Rabinowitz YS. Keratoconus. Surv Ophthalmol 1998;42:297–319.
3. Ugboko VI, Oginni FO, Ajike SO, Olasoji HO, Adebayo ET. A survey of temporomandibular joint dislocation: Aetiology, demographics, risk factors and management in 96 Nigerian cases. Int J Oral Maxillofac Surg 2005;34:499–502. [CrossRef]
4. Kelly DJ, Farrell SM. Physiology and role of intraocular pressure in contemporary anesthesia. Anesth Analg 2018;126:1551–62.
5. Malek S, Reinhold EJ, Pearce GS. The Beighton score as a measure of generalised joint hypermobility. Rheumatol Int 2021;41:1707–16. [CrossRef]
6. Rani S, Pawah S, Gola S, Bakshi M. Analysis of helkimo index for temporomandibular disorder diagnosis in the dental students of Faridabad city: A cross-sectional study. J Indian Prosthodont Soc 2017;17:48–52. [CrossRef]
7. Hirsch C, John MT, Stang A. Association between generalized joint hypermobility and signs and diagnoses of temporomandibular disorders. Eur J Oral Sci 2008;116:525–30. [CrossRef]
8. Sia SL, Chang YL, Lee TM, Lai YY. Temporomandibular joint dislocation after laryngeal mask airway insertion. Acta Anaesthesiol Taiwan 2008;46:82−5. [CrossRef]
9. Street DA, Vinokur ET, Waring GO 3rd, Pollak SJ, Clements SD, Perkins JV. Lack of association between keratoconus, mitral valve prolapse, and joint hypermobility. Ophthalmology 1991;98:170–6. [CrossRef]
10. Woodward EG, Morris MT. Joint hypermobility in keratoconus. Ophthalmic Physiol Opt 1990;10:360–2. [CrossRef]
11. Galperin G, Berra M, Berra A. Keratectasia following laser in situ keratomileusis in a low-risk patient with benign joint hypermobility syndrome. Arq Bras Oftalmol 2014;77:119–21. [CrossRef]