



# Local anesthesia of the temporomandibular joint to reduce pain during mouth opening for dental treatment in a patient with spinal muscular atrophy

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Spinal muscular atrophy (SMA) is an autosomal recessive, severe neuromuscular disorder in which degeneration of alpha motor neurons in the spine progressively weakens and ultimately paralyzes the proximal muscles. It occurs in one per 6,000-10,000 infants, and is a genetic disorder with the second-highest mortality rate worldwide. An 18-year-old male patient with SMA was referred for general anesthesia for difficulty in performing dental treatment due to limited mouth opening caused by temporomandibular joint (TMJ) pain. However, the patient had a high risk of general anesthesia complications, so TMJ pain during mouth opening was reduced through local anesthesia of the TMJ. Fortunately, the anesthesia was successful in reducing pain during mouth opening, enabling the patient to receive dental treatment with an adequate mouth opening.

**Keywords:** Dental care for disabled; Spinal muscular atrophy; Temporomandibular joint disorder.

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Spinal muscular atrophy (SMA) is an autosomal recessive, severe neuromuscular disorder in which degeneration of alpha motor neurons in the spine progressively weakens and ultimately paralyzes proximal muscles [1]. It occurs in one per 6,000-10,000 infants [1], and is a genetic disorder with the second-highest fatality rate worldwide [2,3]. The main clinical symptoms include muscle weakness and muscle atrophy, such as the inability to hold the head up, gait impairment, and inability to lift the arms. Reduced muscle strength induces low ventilation, breathing impairment, reduced coughing ability, and pulmonary collapse, and once pulmonary aspiration occurs from swallowing difficulty and gastroesophageal reflux, pneumonia and respiratory

failure may repeatedly occur [4]. Anesthesia in these patients requires special care, as they are vulnerable to respiratory complications due to muscle weakness, increased sensitivity to nondepolarizing muscle relaxants, and succinylcholine-induced hyperkalemia [5]. For this reason, surgery under spinal anesthesia or local anesthesia is preferred in patients with SMA [6].

The present study reports a case of a patient with SMA who was referred to us for general anesthesia because hyperextension of the mouth closing muscle caused severe pain in the bilateral temporal area and temporomandibular joint (TMJ) during mouth opening for dental treatment. Local anesthesia of the TMJ area enabled successful dental treatment.

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## CASE REPORT

An 18-year-old male patient (height 140 cm, weight 20 kg) was referred to our hospital for dental treatment under general anesthesia with a chief complaint of an inability to open his mouth wide for dental treatment. The patient was diagnosed with spinal muscular atrophy (SMA) at one year of age through muscle biopsy. While he has clear consciousness, he has never walked, as he was unable to move his arms or legs. Since the age of 8, when he suffered aspiration pneumonia, he has been completely bedridden and has been using continuous positive airway pressure (CPAP) during sleep. The patient usually has his neck turned to the left because he has difficulty breathing and sometimes experiences signs of dyspnea when he turns his neck to the right. He underwent two rounds of general anesthesia for scoliosis surgery in 2008 and 2010, and had a history of tracheostomy for dyspnea that occurred in the intensive care unit (ICU) after undergoing general anesthesia for the second time. In a limited manner, the patient was able to voluntarily open his mouth and was able to eat regular food. The patient has been complaining of pain in his TMJ and temporal area during mouth opening for 10 years, which hindered him from receiving treatment for long-term molar pain.

Therefore, he was referred to undergo general anesthesia

for treatment of the molar area. However, we decided to defer general anesthesia due to a high risk of respiratory complications based on his medical history and physical examination, and instead performed local anesthesia of his TMJ area. We determined that the patient had severe pain in the bilateral temporal and TMJ area due to hyperextension of the mouth closing muscle while opening his mouth, and perform local anesthesia to alleviate tension in the TMJ and mouth closing muscle area during dental treatment. At the dental clinic, 1 mL 0.5% levobupivacaine was injected on each side of the TMJ and temporalis fascia before performing oral and maxillofacial surgery. In approximately 10 minutes, the patient reported feeling no pain while opening his mouth, and oral examination and the required dental treatments were performed while monitoring his oxygen saturation levels. During the examination, his #36, #45, and #46 teeth were identified as severely carious with exposed pulp in addition to a severely carious #26 tooth, moderately carious #13, #37, and #42 teeth, and mildly carious #43 tooth. Hence, we extracted #36 and #46, and performed restorative treatment and root canal for #37 and #45, respectively. The patient maintained a limited degree of mouth opening ability (20 mm), and remaining treatments were conducted in subsequent visits without additional local anesthesia of the mouth closing muscle and TMJ.



Fig. 1. One milliliter of 0.5% levobupivacaine each was injected into the right and left TMJ.

## DISCUSSION

Patients with SMA may have various respiratory complications due to respiratory muscle weakness. They are at high risk of aspiration due to swallowing difficulty and reflux, which are critical factors associated with increased mortality [7]. For this reason, anesthesia care for these patients is challenging. SMA patients have high particularly high risks during extubation. According to Bach et al., the success rate of conventional extubation for SMA patients is only about 6%, compared to 85% for protocol extubation, with increased potential for death from tube-related complications [8]. Moreover, patients with neuromuscular diseases may have increased sensitivity to nondepolarizing muscle relaxants and hyperkalemia induced by succinylcholine [5]. Because anesthesia care of SMA patients is very difficult and the patient in the present case had a history of dyspnea following two rounds of general anesthesia, general anesthesia for dental treatment was contraindicated.

In 1999, Granger et al. reported that the biting force of masticatory muscles of patients with SMA was half that of the control group; in addition, these patients also had mouth openings half those of the control group and felt fatigue 30% faster [9]. Furthermore, according to their caregivers, patients with SMA have difficulty in eating tough or hard food, take a long time to eat, and require breaks during eating. These factors not only have adverse effects in managing oral hygiene, but also hinder dental treatment. The patient in the present study required treatment for multiple caries, but was referred to us because he could not open his mouth due to pain. The patient had highly limited mouth opening and complained of pain in the temporal and TMJ area on both sides when he tried to open his mouth wider. He reported that he typically ate regular meals, which may have resulted in muscle fatigue, leading to local muscle pain in the masticatory muscles, which in turn reduced the range of mandibular motion as a protective co-contraction. Another cause might have been that the patient did not

have to open his mouth wider than required for eating or talking along with limited mouth opening in SMA patients, and such limited mandibular movement might have caused myostatic contracture in the mouth opening muscles. In such cases of temporomandibular disorder (TMD), local anesthetics could be useful. If the cause of the pain lies in the muscles or joints, injecting local anesthetics in the source could eliminate the pain and enable a definitive diagnosis [10]. In 2013, Nascimento et al. reported that 0.5% Bupivacaine without vasoconstrictor during conduction anesthesia of the auriculotemporal nerve significantly reduced pain [11]. Hence, we performed anesthesia of the auriculotemporal nerve using 0.5% bupivacaine. The patient reported not feeling pain when opening his mouth after receiving the local anesthesia, and thus was able to undergo dental treatment without general anesthesia.

Many neurologists do not believe that SMA, a lower motor neuron disease, is associated with TMD because the masticatory muscles are controlled by the trigeminal motor nucleus [12,13]. However, many cases and studies report that TMD and limited mouth opening are common complications experienced by patients with SMA [9, 14-16]. Therefore, increasing mouth opening via local anesthesia administered at the source of pain during mouth opening could be an option to consider in SMA patients with chronic TMD that hinders dental treatment.

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