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A case of sublingual ranula that successfully responded to micro-marsupialisation under COVID-19 infection: A case report

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ARTICLE INFO	A B S T R A C T
Keywords: Ranula OK-432 Micro-marsupialisation COVID-19	Ranula is a mucous retention cyst caused by the extravasation of mucus from the sublingual gland. We present a case of sublingual ranula that was successfully treated with micro-marsupialisation under COVID-19 infection. The patient was a 17-year-old Japanese male suffering from a sublingual ranula that did not improve after several rounds of puncture-aspiration therapy. The patient underwent OK-432 injection therapy under hospitalisation. However, the swelling worsened. Thus, micro-marsupialisation was subsequently performed. After micro-marsupialisation, the lesions flattened out, but 14 days after treatment, the patient was found to have asymptomatic COVID-19 infection when he underwent polymerase chain reaction testing as a close contact person. Simultaneously, the lesion re-swelled and became painful, so non-steroidal anti-inflammatory drugs were prescribed. The next day, the sutures spontaneously detached, viscous saliva and blood overflowed, and the

1. Introduction

Ranula is a cystic lesion in the floor of the mouth that develops due to the extravasation of mucus from the sublingual gland. Various treatment modalities have been reported, including aspiration, marsupialisation, OK-432 injection, ranula excision, and sublingual gland plus ranula excision. However, there is no standard treatment due to the different complications and recurrence rates [1].

We present a rare case of sublingual ranula that successfully responded to localised injection treatment with OK-432 (Picibanil®, Chugai Pharmaceutical Co, Tokyo, Japan) and micro-marsupialisation under COVID-19 infection.

2. Case report

A 17-year-old Japanese male was referred to our department with repeated painful swelling of the floor of the mouth despite three aspiration treatments over a two-month period.

Under hospitalisation, the patient underwent OK-432 injection therapy. However, the swelling worsened postoperatively. Thus, micro-marsupialisation, according to Sandrini et al.'s technique [2], was selected and performed 3 months after OK-432 injection (Supple Fig. 1).

Over the course of a few days after the microsurgical procedure, the lesion gradually flattened, and the patient progressed well. Fourteen days after the procedure, he underwent polymerase chain reaction (PCR) testing due to being in close contact with a COVID-19 patient. Although he was asymptomatic, COVID-19 was detected. Seventeen days after the procedure, the patient consulted us about re-swelling of the ranula and pain, and non-steroidal anti-inflammatory drugs were prescribed. The next day, the patient experienced spontaneous suture dehiscence and much discharge of viscous saliva mixed with haemorrhage, and the ranula gradually disappeared. Twenty-four days after the procedure (10 days after being COVID-19 positive), the patient was discharged after a negative PCR test result. At that time, the ranula had resolved, but the patient remained coughing and fatigued, which were sequelae of the COVID-19. Six months have passed since the treatment, and no evidence of recurrence of the ranula has been noted to date, but the sequelae of the COVID-19, cough and mild malaise, remained (Supple Fig. 1).

ranula disappeared. Micro-marsupialisation is effective and useful even if the patient has a COVID-19 infection.

3. Discussion

In the present case, the ranula reappeared swollen and inflamed after the COVID-19 infection, leading to a healing process with micro-

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marsupialisation. Interestingly, swelling in the floor of the mouth has rarely been reported as a potential manifestation of COVID-19 [3]. Since angiotensin-converting enzyme 2, the COVID-19 receptor, has been reported to be highly expressed in the oral cavity [4], it is possible that a strong inflammatory reaction occurred at the same site. Therefore, the remaining ranula may have been inflammatory and re-swollen with severe pain and expelling saliva mixed with haemorrhage. Consequently, micro-marsupialisation was successful under COVID-19 infection, and the ranula disappeared.

Ranulas are usually treated by surgical approaches, including sublingual gland excision, due to their high recurrence rate. However, iatrogenic complications, such as injury of the lingual nerve and Wharton's duct, are major concerns. Although OK-432 injection may be relatively safe and effective, the drawback is that multiple doses are often required before the healing occurs [5]. Furthermore, OK-432 induces inflammation, which results in painful swelling. As for the marsupialisation used in this case, there are many reports recommending it over sublingual gland excision because of its simplicity and effectiveness [2,6–10].

In this case, although re-swelling was observed after the COVID-19 infection, the patient spontaneously healed without the need for subsequent hospital visits, which reveals the effectiveness of the micromarsupial technique as a first-line treatment for orally localised ranula in situations where COVID-19 infection is widespread.

Ethics statement/confirmation of patient's permission

The institutional review board has confirmed that no ethical approval is required. Patient and patient's family signed a written informed consent for using patient's data and information's, including photos, for publication.

Declaration of competing interest

All the authors have no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.adoms.2022.100309.

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