Abstract A 36-year-old man was admitted who had a 2-month history of swelling of the right submandibular area and the right side of the mouth floor. He had undergone operations for right plunging ranula with a lesion on the floor of the mouth twice 7 years ago. Under fluoroscopic guidance, the contents of the cyst were aspirated as much as possible and injected with the same amount of OK-432 solution (Picibanil 0.1 mg; 10 ml) twice with 3-week intervals. Examination after 6 weeks showed that the cystic mass seen before therapy had disappeared completely, and no recurrence was encountered after 1 year. We here report a case in which a successful sclerotherapy with OK-432 for recurrent plunging ranula after surgery was performed.

Keywords OK-432 (Picibanil) · Recurrent plunging ranula

Introduction

Plunging ranula is a relatively uncommon phenomenon, which represents a mucus escape reaction occurring because of the disruption of the sublingual salivary gland. It may appear as a submandibular mass without visible intraoral involvement. Surgical excision is the first choice of therapy [3]. However, insufficient surgery and frequent recurrence because of the lack of the cystic capsule and the risk of damage to the surrounding vital structures, particularly nerves and blood vessels, have been reported. Nonsurgical therapies have been attempted to avoid surgery-related complications [4].

Recently, OK-432 (Picibanil), a sclerosing agent, has been reported to be highly effective and to have minimal side effects in the treatment of cystic lymphangioma and plunging ranula [3]. This report describes the successful treatment of postoperative recurrent plunging ranula by intralesional injection of OK-432 (Picibanil).

Case report

A 36-year-old man who had a 2-month history of swelling of the right submandibular area and the ipsilateral floor of the mouth visited our department on 1 August 2000. At the physical examination, a 5x5-cm-sized, mobile and fluctuating cystic mass in the submandibular area with a previous operative scar was detected (Fig. 1). Seven years ago, the man had been operated on transcervically for the same lesion twice. The histologic diagnosis of a previous operative specimen was consistent with plunging ranula. A computed tomography scan of the neck showed a non-enhanced, poorly demarcated and uncapsulated cystic mass of low density in the right parapharyngeal space, and the right submandibular gland was absent because of previous surgery (Fig. 2). From these find-
ings, a diagnosis of recurrent plunging ranula was given, and sclerotherapy was planned using OK-432 (Picibanil, Chugai Pharmaceutical Co., Japan). The dosage and method of intralesional injection was used according to Ogita’s description [5, 9]. Briefly, OK-432 solution was prepared by diluting 0.1 mg of OK-432 in 10 ml of normal saline with contrast. Under fluoroscopic guidance, the cystic mass was punctured, and its contents were aspirated as much as possible, and the same amount of OK-432 solution was injected (Fig. 3). The following day, the patient visited the emergency room with a high fever and painful swelling of the lesion. The symptoms were improved by the administration of antipyretics and hydration. As a sufficient response was not observed, the second injection was given after 3 weeks. There were no previous side effects. After another 3 weeks, the swelling of the right submandibular area and floor of the mouth had disappeared. No recurrence of the ranula was observed during the subsequent 1 year of follow-up.

Discussion

Plunging ranula occurs in 10% of all ranula cases. It needs to be distinguished from lymphangioma, although surgery is the first-choice therapy for both of these. Frequent recurrences of the disease because of insufficient surgery have been reported, and various therapies have been designed in addition to surgery. Various sclerosing agents, such as iodine, ethanolamine oleate, alcohol, bleomycin, tetracyclin and cyclophosphamide, have been used since the time when sodium morrhuate was used for sclerotherapy in 1933 [2]. Their results were variable and had numerous other local and systemic side effects [1, 10]. In 1987, Ogita et al. [9] reported good results in the treatment of lymphangioma using OK-432. Thereafter, the effectiveness of OK-432 was confirmed by the treatment of lymphangiomas in children and of unresectable and recurrent lymphangiomas [6, 7]. In 1994, Ikarashi et al. reported cases of plunging ranula treated by OK-432 therapy [3]. In our case, the lesion was poorly demarcated from the vital structures and could be seen adhering to the surrounding tissues on computed tomography scan of the neck; recurrence was highly anticipated. We decided to treat the lesion using OK-432, and the result was satisfactory.

The working mechanism of OK-432 in the lesions is that it immediately evokes inflammation and the infiltration of the inflammatory cells into the cystic spaces. After the injection of OK-432, there is extensive production of cytokines, including interleukin-6 and tumor-necrotic factor (TNF) [8]. These cytokines increase the endothelial permeability. The accelerated lymph drainage and increased lymph flow lead to shrinkage of the cystic spaces. Furthermore, NK cell activity was potentiated by OK-432, and helper- and killer-T cells increased in number, which suggests that there is some relationship between these changes and the shrinkage of the cyst [8]. The response does not appear to extend beyond the cystic cavity and does not lead to scar formation on the overlying skin. As for side effects, transient fever and swelling that are universally noted in the first few days after injection are treated easily with antipyretics. Because of the possibility of upper airway obstruction resulting from post-injection swelling of the peritracheal lesion and of allergic reactions to penicillin G, a careful selection of indications is required [4, 11].

In this patient, the intralesional injection of OK-432 has been an effective method for the treatment of recurrent plunging ranula with an avoidance of surgical complications.

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References