Huge Lymphangioma of the Tongue: A Case Report

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Lymphangioma of the tongue is relatively rare and may cause facial structural deformity. Using a combination of a V-shaped and central resection, we successfully treated a 6-year-old girl who had massive lymphangioma of the tongue. Postoperatively, her tongue was located completely within her mouth with good cosmetic results. Sensory and motor nerves to the tongue appeared to be intact. Her speech was also improved. [Asian J Surg 2003; 26(4):228-30]

Introduction

Lymphangiomas are congenital benign tumours of the lymphatic system, and 50% to 75% are located in the head and neck.1 They may arise in the tongue, and are a common cause of macroglossia in children. Lymphangiomas of the tongue (LTs) do not regress spontaneously, and they are almost always of the cavernous type.2 The lesions are not tender or painful. However, inflammation from trauma or infection may result in sudden enlargement and severe pain. LT may cause gross structural deformities of the face and interfere with speech and swallowing.2 We report a case of massive LT, in which surgical resection was successfully performed, leading to good cosmetic and functional results.

Case report

A girl born with macroglossia was diagnosed with LT and managed conservatively at a district hospital. When she was 5 years old, the tongue suddenly enlarged after an episode of infection (Figure 1). Following this, she had many problems; the tongue could not be withdrawn into the oral cavity, the mouth could not be closed, and saliva dribbled from the corners of her mouth. She was eating only soft food with liquid and enteral nutritional supplementation. Her sensation of taste was intact and her speech was not severely affected. There were no other associated anomalies. At the age of 6, she was

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Figure 1. Massive lymphangioma in the tongue: A) frontal view; B) lateral view.
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transferred to our hospital for resection of the lesion. Magnetic resonance imaging (MRI) showed that the LT was localized within the tongue, with no extension into the pharynx or neck (Figure 2).

The Köle technique, which is a combination of V-shaped resection and central resection, was used under general nasotracheal anaesthesia. Approximately 6 × 5 cm of the tumour was resected by partial glossectomy in the shape of a boat’s keel (Figure 3), and care was taken to minimize injury to the VII, IX and XII cranial nerves. After resection, the tongue was relatively normal in size and shape, although some lymphangiomaticous tissue remained in each side of the tongue. The postoperative course was uneventful. Histopathological examination of the resected specimen showed the presence of many cavernous lymphatic spaces within the lamina propria and the underlying muscle.

The patient was discharged 2 weeks after surgery, with a normal-looking tongue and marked improvement in speech (Figure 4). At follow-up 6 months after surgery, her tongue was located completely within her mouth, with good cosmetic results. Tongue sensory and motor functions were intact and her speech was continuing to improve. She has a mandible deformity and anterior open bite secondary to chronic untreated macroglossia and will probably require orthodontic treatment.

Discussion

The goal of treatment of LT is to obtain good cosmesis, prevent facial deformity, and prevent speech problems. Surgical resection, radiation therapy, cryotherapy, electrocautery, sclerotherapy, steroid administration, embolization, ligation and laser surgery are treatment options for LT. Lymphangioma is resistant to medical treatment. Radiotherapy is effective in the early stages, but the tumour recurs in many cases. Although the sclerosing agent OK-432 is effective for cystic lymphangioma, it is not usually effective in the cavernous type. Because of this, surgical resection appeared to be the most effective treatment for LT in this case.

Partial glossectomy of the tongue has been reported to be an effective treatment for LT. Typical glossectomy can be roughly divided into three procedures: V-shaped resection, bilateral marginal resection, or central resection. Taste is detected by the tongue, the soft palate, the pharynx and the buccal mucosa. Although the whole tongue can detect taste, most of the taste buds are located on the sides of the tongue. Thus, when the tongue is resected, it is desirable to leave the sides intact. To limit injury to taste and motor functions, a central resection is preferable; however, satisfactory resection

Figure 2. Magnetic resonance image shows that the lymphangioma is localized within the tongue (arrows). There is no extension into the pharynx and neck.

Figure 3. Intraoperative photographs: A) planned incision; B) resection of the tongue; C) resected lymphangioma of the tongue; and D) approximation of the remaining tongue.
Surgical resection of LT is indicated to prevent orthodontic, phonetic, psychological and cosmetic problems. If the mouth cannot close due to an enlarged tongue, this will cause occlusal imbalance, mandible deformity and delay of speech development. It will be difficult to treat these problems after a growth spurt starts. Thus, surgical reduction of the tongue is recommended before the anterior teeth have been completely replaced by adult teeth (around 7 years of age). In our case, psychological problems were resolved and quality of life improved dramatically after surgery. However, careful long-term follow-up is required because mandible deformity and anterior open bite still remain, and LT may recur from the residual lesions.

References