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Case Report

Masseter muscle hypertrophy: A case report

Masayuki Tsuneki[a], Satoshi Maruyama[a], Manabu Yamazaki[b], Kanae Niimi[c], Tadaharu Kobayashi[c], Hideyoshi Nishiyama[d], Takafumi Hayshi[d], Jun-ichi Tanuma[a,b,*]

[a] Oral Pathology Section, Department of Surgical Pathology, Niigata University Hospital, Niigata 951-8520, Japan
[b] Division of Oral Pathology, Niigata University Graduate School of Medical and Dental Sciences, Niigata 951-8514, Japan
[c] Division of Reconstructive Surgery for Oral and Maxillofacial Region, Niigata University, Graduate School of Medical and Dental Sciences, Niigata 951-8514, Japan
[d] Division of Oral and Maxillofacial Radiology, Niigata University Graduate School of Medical and Dental Sciences, Niigata 951-8514, Japan

ABSTRACT

Masseter muscle hypertrophy is defined as the unilateral or bilateral expansion of the masseter muscle, and little is known about its etiology. Here we report a case of masseter muscle hypertrophy in a female patient in her 20 s who complained of facial asymmetry. Because of the hemifacial overgrowth of the right-side maxillofacial region from birth, she was thought to have congenital hemifacial hyperplasia. After orthodontic treatment and osteotomy, the patient underwent debulking of the right masseter muscle. Histologically, the surgical specimens exhibited thick masseter muscle fibers arranged irregularly. Masseter muscle cells exhibited diameters in the range of 20–60 μm.

1. Introduction

Masseter muscle hypertrophy is characterized by an asymptomatic enlargement of unilateral or bilateral masseter muscles. Although the etiology is unclear, it may be a result of hyperfunction caused by bruxism [1–3]. Congenital hemifacial hyperplasia was recognized by Gesell in 1927 and was described as “essentially a developmental anomaly antedating birth and arising in some way as a partial deflection of the normal process of birth”. The asymmetric enlargement of hemifacial hyperplasia may be manifested in the muscles, vascular system, and nerves [4]. We report a case of a female patient in her 20 s with masseter muscle hypertrophy in hemifacial hyperplasia. In this report, we show the case of a 20’s female patient with masseter muscle hypertrophy in hemifacial hyperplasia.

2. Case report

A 24-year-old female patient was referred to Niigata University Medical and Dental Hospital with a request for the treatment of facial asymmetry present since birth (Fig. 1). She had no family history of facial asymmetry. Because of the hemifacial overgrowth of the right-side maxillofacial region from birth, she was thought to have congenital hemifacial hyperplasia.

The patient’s face was asymmetrical with an enlargement of the right side including the malar, maxillary, and mandibular regions (Fig. 1). Malocclusion was evident, with an anterior open bite and distortion of the dental arches. The patient’s skin and hair appeared normal. After orthodontic treatment and osteotomy, the patient underwent debulking of the right masseter muscle.

We performed a morphometric examination for measuring the diameter of the muscle fibers in the masseter muscles in at least three of the 30 muscle cells. Normal masseter muscle fibers are arranged irregularly with a diverse range of diameters less than 20 μm (average diameter 12–13 μm) [1–4]. The normal masseter muscle fibers specimens used for hematoxylin and eosin staining and immunostaining in this study were sourced from other patients with non-inflammatory diseases. Histologically, the surgical specimens exhibited thick masseter muscle fibers arranged irregularly with a diverse range of diameters

* Corresponding author at: Division of Oral Pathology, Niigata University Graduate School of Medical and Dental Sciences, 2-5274, Gakkocho-dori, Chuo-ku, Niigata 951-8514, Japan.
E-mail address: tanuma@dent.niigata-u.ac.jp (J.-i. Tanuma).

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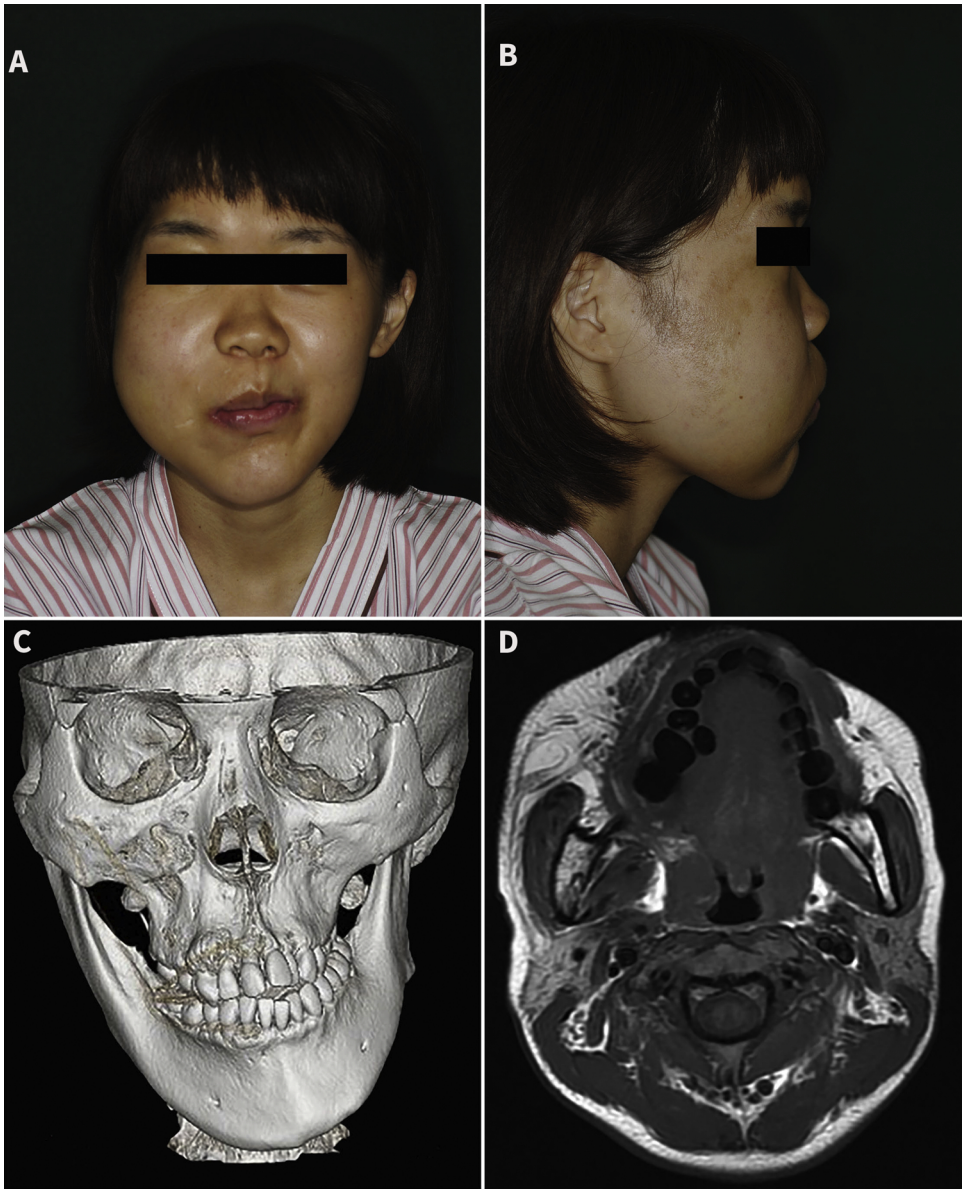

Fig. 1. Masseter muscle hypertrophy in hemifacial hyperplasia.
(Lower left: CT image; Lower right: MRI)

from 20 to 60 μm (Figs. 2 and 3). Immunohistochemically, calcineurin A was preferentially localized in the cytoplasm of the hypertrophic masseter muscle cells when compared with normal cells (Fig. 4) [5].

3. Discussion

Masseter muscle hypertrophy is a relatively rare and benign enlargement of unilateral or bilateral masseter muscles. This asymptomatic persistent muscle enlargement may be initiated by bruxism, clenching, or heavy gum chewing [3,6]. However, idiopathic and congenital hypertrophy of the masseter muscle is a rare disorder of unknown etiology [6].

Hemifacial hyperplasia is a rare congenital developmental anomaly exhibiting significant unilateral enlargement of the maxillofacial hard and soft tissues [4,7]. Women are more commonly affected than men, with the right side predominantly affected as in this case. Several etiological factors (e.g., heredity, chromosomal abnormalities, altered intrauterine development, endocrine dysfunction, and vascular/

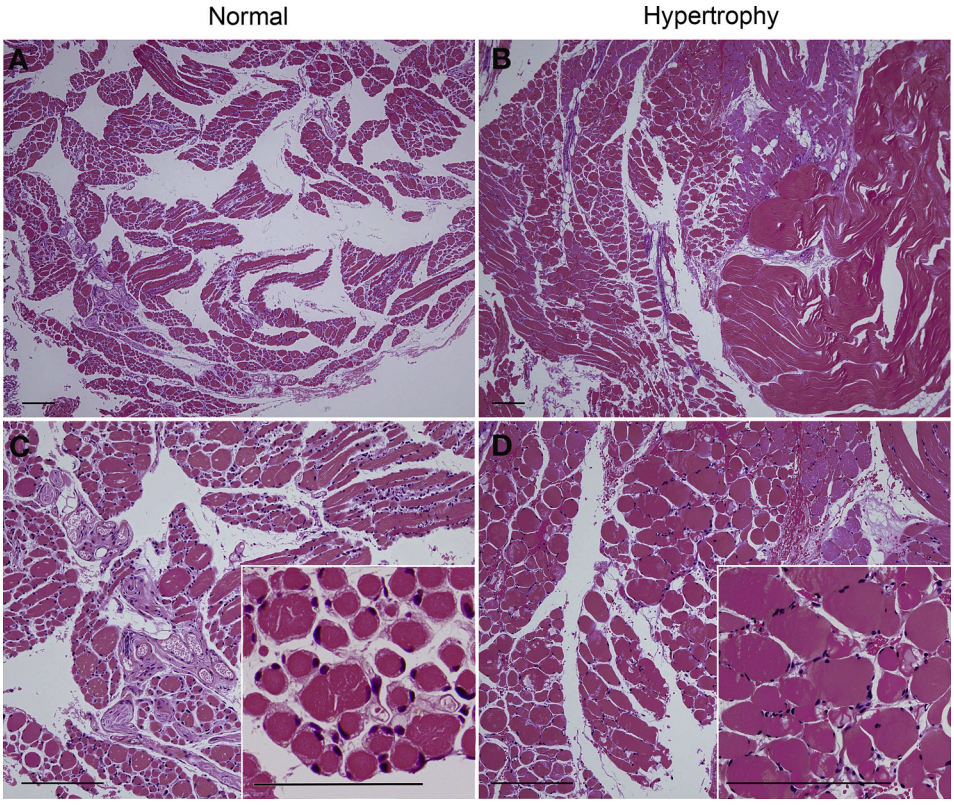

Fig. 2. Histopathology of masseter muscle hypertrophy. (Scale bars: 200 μm).

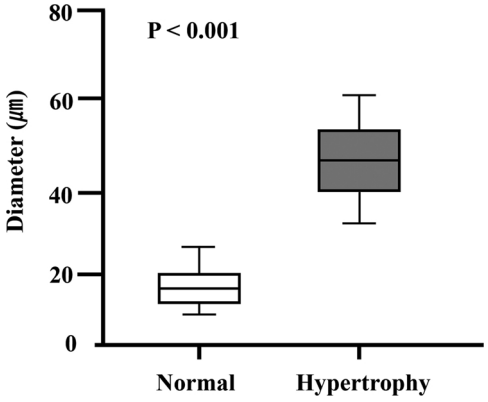

Fig. 3. Diameters of masseter muscle cells.

lymphatic abnormalities) remain implicated in this condition; however, no single factor has been directly related to its manifestation [8].

In this report, we describe a case of masseter muscle hypertrophy in congenital hemifacial hyperplasia. Hemifacial hyperplasia is associated with a diverse range of abnormalities, such as enlargement of the hard and soft tissues on the affected side. Frequently, the tongue exhibits unilateral enlargement with thickening and hypertrophy [9]. In the present case, the tongue was within the normal size range, but an anterior open bite, distortion of the dental arches, and unilateral masseter muscle hypertrophy were evident.

In the literature, histological investigation of specimens from patients with masseter muscle hypertrophy have revealed pronounced hypertrophy with an increase in the muscle fiber diameter to approximately 50 μm, which is more than twice to three times as large as normal muscle fibers [3,10,11]. These findings are consistent with those from our case study.

Studies using an animal model (bite-opening rats) proposed that calcineurin signaling [12], which is a calcium/calmodulin-regulated protein phosphatase which acts on the transcription factors of the nuclear factor of activated T cells (NFAT) family is an important molecular mechanism driving masseter muscle hypertrophy [13–15]. In the present case, calcineurin A was preferentially expressed in the cytoplasm of hypertrophic masseter muscle cells when compared with normal masseter muscle cells. Thus, calcineurin A may play a pivotal role in the development of masseter muscle hypertrophy in congenital hemifacial hyperplasia.

In conclusion, masseter muscle hypertrophy is a rare disorder that can develop in congenital hemifacial hyperplasia. Calcineurin A signaling is a potential driver for developing masseter muscle hypertrophy.

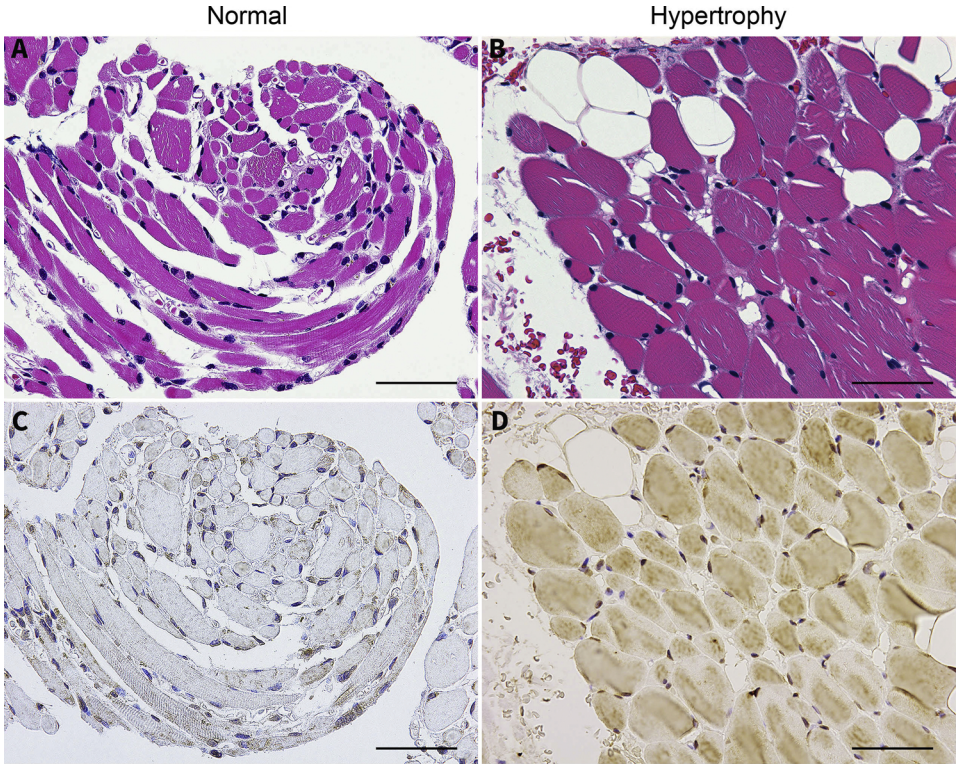

Fig. 4. Calcineurin A expression in masseter muscle cells. (Scale bars: 50 μm).

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