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Intramuscular Cavernous Hemangioma of the Masseter Muscle in Child and Adolescent

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교근에서 발생한 근육내 혈관종 2예

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이재운 · 조완석 · 김승범 · 이동훈

= 국문초록 =

근육내 혈관종은 주로 몸통과 사지에 발생하며, 두경부 영역에서의 발생은 드문 것으로 알려졌다. 저자들은 교근에서 발생한 2예의 근육내 혈관종을 치험하였기에 문헌 고찰과 함께 보고하는 바이다. 임상의사들은 특별한 원인 없이 진행되는 이하선 부위의 종창을 주소로 내원하였을 때, 교근에서 발생한 근육내 혈관종의 가능성도 염두 해두어야 한다.

중심 단어 : 근육내 · 혈관종 · 교근.

Introduction

Intramuscular hemangiomas occur most often in the trunk and extremities, and are uncommon tumors of the head and neck.[1] The rarity, unfamiliar presentation, and deep location of intramuscular hemangioma, led to an accurate preoperative diagnosis in fewer than 8% of cases.[2,3] We reported 2 cases of intramuscular hemangioma of the masseter muscle in a child and adolescent, which were diagnosed preoperatively and successfully treated by surgery without complications.

Case Report

1. Case 1

A 7-year-old boy was referred to our hospital with a complaint of painless swelling of the right cheek. The patient's parents first noticed the swelling 6months prior, and it had gradually increased in size. They denied any history of trauma and operation.

A 3 cm sized round, mildly compressible mass was palpable over the right parotid region, on physical examination. No pulsations, bruits or change in skin color was noted. Fine-needle aspiration cytology(FNAC) showed some scattered lymphoid cells and histiocytes in a bloody background. Computed tomography(CT) scan of the neck revealed a 3.5 cm, poorly defined heterogeneously enhancing lesion located within or adjacent to the right masseter muscle(Fig. 1). The lesion had low T1-weighted and high T2-weighted signal intensities on Magnetic resonance imaging(MRI), within the right parotid gland with extension to right masticator space (Fig. 1).

The patient was diagnosed with an intramuscular hemangioma of the right masseter muscle, based on these observations. A standard 'lazy S' cervico-mastoid-preauricular surgical incision was made. An intraoperative facial nerve monitor was used. The intraoperative findings were that of a

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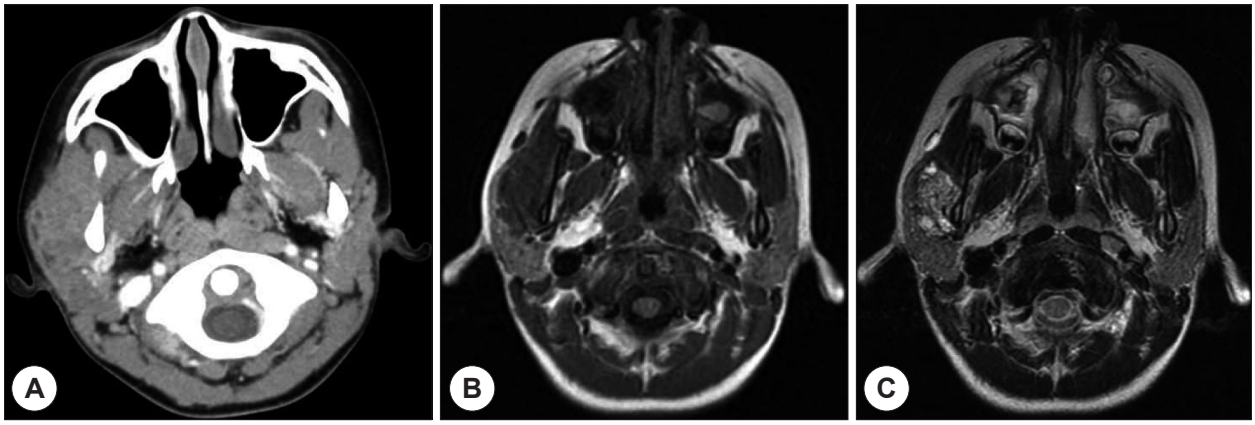

Fig. 1. Axial CT scan of the neck with enhancement(A) shows a 3.5 cm mass in the masseter muscle anterior to the right parotid gland. MRI reveals about 2.8×2.3×2.1 cm heterogeneous enhancing mass in right parotid gland and masseter muscle with extension to masticator space. The lesion had low T1-weighted(B) and high T2-weighted(C) signal intensities.

Fig. 2. Intraoperative view shows that intramuscular hemangioma was removed totally with preservation of the facial nerve.

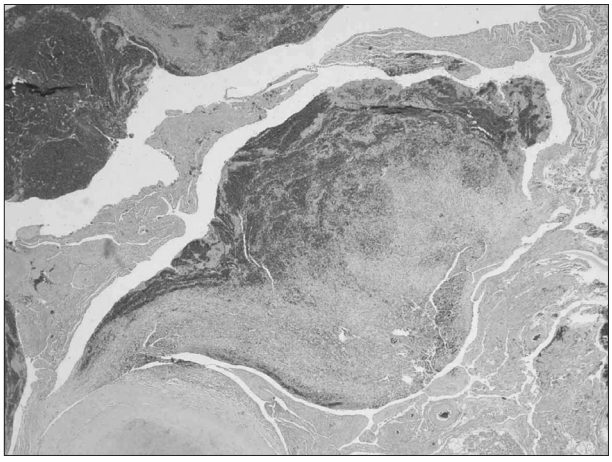

Fig. 3. Histopathologic examination shows large dilated vessels lined by flattened enothelium and thrombus formation with hemosiderin pigments(hematoxylin and eosin, ×20).

highly vascular tumor involving the masseter muscle and a normal parotid gland superficial to it. No large vascular connection and invasion adjacent structures were noted. The tumor and masseter muscle were removed completely with preservation of the facial nerve(Fig. 2). Pathological examination of the lesions revealed cavernous hemangioma(Fig. 3). The postoperative course was uneventful. There was no evidence of recurrence at the 6 year follow-up.

2. Case 2

A 17-year-old girl was referred for evaluation and management of progressive, painless swelling of the left cheek. The swelling had been present since 4 years ago, and had gradually increased in size. No prior history of trauma, dental problems, operation, or other medical problems were noted.

Physical examination showed a roughly 4cm soft, mobile, mildly compressible mass in the left parotid region. No pulsations, bruits or change in skin color was noted. FNAC showed a suspicious inflammatory lesion. CT scan of the neck revealed a 4×3 cm, well defined soft tissue density mass located within or adjacent to the masseter muscle(Fig. 4). MRI on T1-weighted image with enhancement showed a 4×2.5 cm well-marginated, heterogeneously enhancing lesion in the left masseter muscle(Fig. 4).

Preoperative diagnosis was hemangioma of the masseter muscle and surgery was scheduled. The surgery was performed with a standard 'lazy S' cervico-mastoid-preauricular incision using intraoperative facial nerve monitor. A dark colored vascular mass was found within the masseter muscle, anterior to the parotid gland. No large vascular connection and invasion surrounding tissue were noted. The branches of the facial nerve were identified and preserved, and the tumor was completely removed with a margin of normal surrounding muscle(Fig. 5). Histopathologic examination confirmed a cavernous hemangioma(Fig. 6). The postoperative course was uneventful. There was no evidence of recurrence at the 4 year follow-up.

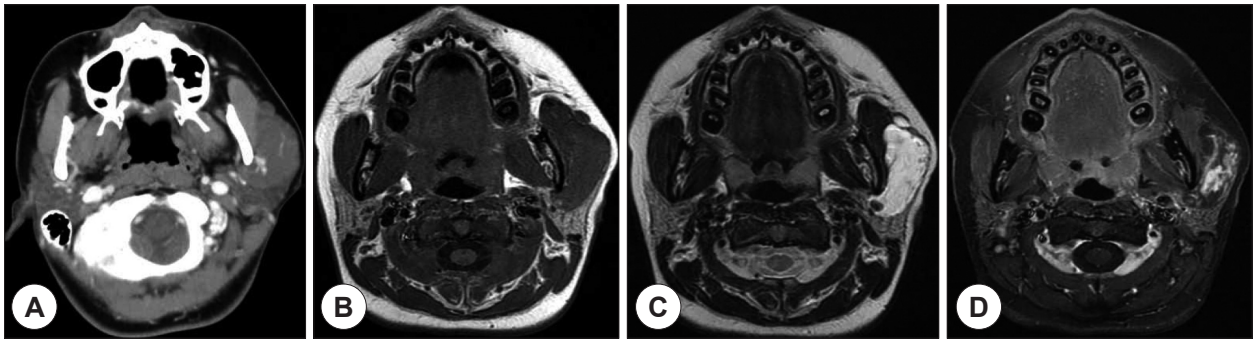

Fig. 4. Axial CT scan of the neck with enhancement(A) shows a 4×3 cm, well defined soft tissue density mass located within or adjacent to the masseter muscle. On MRI, the lesion had low T1-weighted(B) and high T2-weighted(C) signal intensities. T1-weighted image with enhancement(D) showed a 4×2.5 cm well-marginated, heterogeneously enhancing lesion in the left masseter muscle.

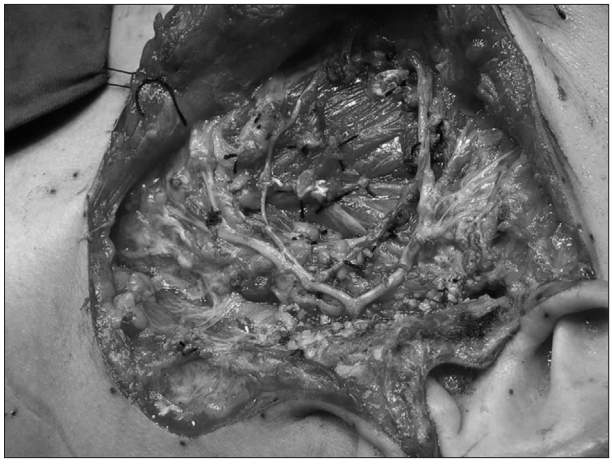

Fig. 5. Intraoperative view shows that the branches of the facial nerve were identified and preserved, and the tumor was completely removed with a margin of normal surrounding muscle.

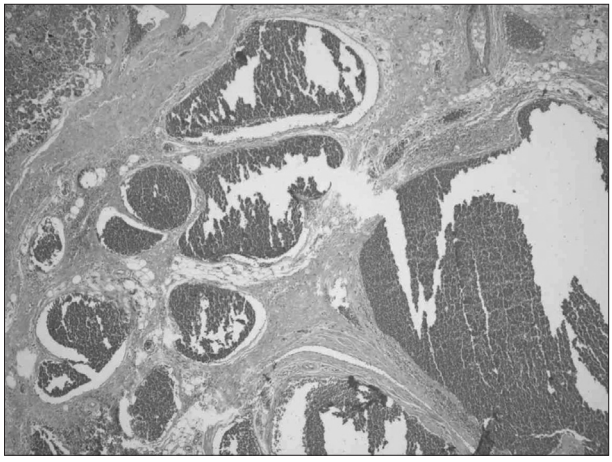

Fig. 6. Histopathologic examination shows large dilated vessels lined by flattened enothelium(hematoxylin and eosin, ×40).

Discussion

Intramuscular hemangiomas are rare, benign neoplasms that represent 0.8% of all benign vascular neoplasms.[4] Approximately 15% of the intramuscular hemangioma are manifested in the head and neck, with masseter muscle being the most common site, followed by the trapezius and sternocleidomastoid muscles, respectively.[2,3] Intramuscular hemangiomas occur most frequently in the third decade.[5] We reported patients who were younger than previously reported, however we established the exact preoperative diagnosis by a presumptive diagnosis of intramuscular hemangioma, followed by radiologic examinations.

Congenital etiology of intramuscular hemangiomas has been widely accepted by most authors.[2,6] Traumatic and hormonal influences have been suggested and may contribute to the etiology or growth spurts.[2,6,7] However, the patients in this study had no history of trauma, medication, or medical problems.

Histologic subtyping is the most widely accepted classification of hemangiomas.[2,7] Allen et al. classified them as large vessel(>140 mm in diameter, cavernous), small vessel(<140 mm in diameter, capillary), and mixed vessel types.1 Capillary type hemangiomas account for 50% of all intramuscular hemangiomas, and occur more frequently in the head and neck region. Cavernous and mixed types occurred more frequently in the trunk and lower limbs.[2,7] However, the unique clinical finding in this study was intramuscular cavernous hemangioma in the head and neck region.

Intramuscular hemangiomas generally present as progressively enlarging masses.[6-8] The most common clinical presentation is a mass with associated pain in 50 to 60% cases, due to rapid enlargement and compression of adjacent structures.[6-8] Muscular fibers surrounding the hemangioma can be responsible for the lack of bruits, thrills, and compressibility in intramuscular hemangiomas, unlike other vascular malformations.[6,7] Overlying skin color change is also rare.[6] Both patients had no pain, bruits, thrill, and change in skin color.

MRI is thought be the most helpful and accurate investigation in the diagnosis of intramuscular hemangioma.[2,6-8] Intramuscular hemangiomas typically show low signal intensity in T1-weighted images and high signal intensity in T2-

weighted images, as seen in our cases.[6] Color doppler sonography is useful to demonstrate the vascular structures in and around muscles and to evaluate the pathological changes like fibrosis and calcifications.[1] Plain X-ray, angiography, CT scan, and FNAC may not be specific and diagnostic.[2,7] Angiography is also probably not indicated unless there is a high preoperative suspicion of large vascular connections to tumor, as evidenced clinically by strong pulsations, bruits and thrills.[2,3] However, angiography is less effective for smaller, low-flow hemangiomas.[9] Rare intramuscular hemangiomas are commonly confused with other benign or malignant lesions, such as cysts, lymphangioma, rhabdomyosarcomas, masseteric hypertrophy, and schwannomas.[7,8] The preoperative information provided by MRI is much more useful clinically to rule out other diseases.[2]

The management of intramuscular hemangioma should be individualized according to the extent of the tumor, tumor growth rate, patient age, and cosmetic considerations.[7-9] The many treatment modalities include cryotherapy, steroid and sclerosing agent injection, embolization, and radiation therapy. However, the treatment of choice for intramuscular hemangioma of the masseter muscle is surgical excision of the lesion and affected muscle, because of infiltrative nature of intramuscular hemangioma and prevention of recurrence.[6,7-9] The surgical approach for intramuscular hemangioma of the masseter muscle should allow safe wide excision with preservation of the facial nerve. We used a standard 'lazy S' cervico-mastoid-preauricular incision, and successfully removed the tumors without facial nerve palsy. This approach achieves excellent exposure and allows identification of the facial neve branches before removal of the lesion.[3] Local recurrence occurred in 9% of intramuscular hemangioma.[6] We found no recurrence at the 6 and 4 year follow-up, because of wide excision with normal muscle margins.

In conclusion, intramuscular hemangioma of the masseter muscle is a rare, benign neoplasm of the head and neck, and difficult to diagnose preoperatively. The patient, who develops a progressive swelling on the cheek without specific causes, should alert the clinician of possible intramuscular hemangioma of the masseter muscle on differential diagnosis, even in children. We recommend performing the MRI scan for accurate diagnosis of intramuscular hemangioma. Surgical excision of the lesion and affected muscle with preservation of facial nerve is essential on diagnosis of intramuscular hemangioma.

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