Intramuscular haemangioma of mylohyoid muscle: A case report

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Abstract

Objective To report a case of intramuscular haemangioma (IMH) with a rare presentation in the mylohyoid, with emphasis on the clinical appearance, and histologic characteristics of the lesion.

Method Case report and review of the literature.

Conclusion Neck swellings can often present a diagnostic dilemma, with a wide preoperative differential diagnosis. IMH are rare benign haemangiomas occurring within the skeletal muscle. They account for approximately 1% of all haemangiomas. These are uncommon in the head and neck region and occur most frequently in the trunk and extremities. In the head and neck, masseter and trapezius are the most common sites involved. Intramuscular haemangioma is seldom diagnosed preoperatively, perhaps due to unfamiliarity with this uncommon lesion and non-specific clinical findings.

Keywords Mylohyoid · Intramuscular haemangioma · Benign · Neck swelling

Introduction

Haemangiomas are mostly tumors of infancy, occurring more on mucosal and cutaneous surfaces. Approximately 50% occur in the first decade, with 94% occurring before the age of 30 years, with no gender predilection [1]. Although >1% of haemangiomas occur in skeletal muscles, 15% of IMH arise in the head and neck region [2]. The masseter and the trapezius muscles represent the most common site of involvement in the head and neck area, mylohyoid being a very uncommon location. To our knowledge only a few cases of IMH of the mylohyoid have been reported in medical literature [3].

We report a case of IMH of the mylohyoid presenting as an unusual neck swelling. The problems involved in the differential diagnosis and treatment are discussed.

Case report

A 21-year-old-female patient presented to the Department of ENT and Head and Neck Surgery at St. John's Medical College Hospital, a tertiary referral center in South India with a history of persistent neck swelling of 5 months duration. Examination revealed a painless, non-compressible swelling, about 3 × 2 cm in the submental region above the hyoid. The swelling was soft in consistency and moved on deglutition and with protrusion of tongue. Ultrasonographic examination showed a submental hypoechogenic collection. Fine needle aspiration cytology (FNAC) performed, resulted in a hemorrhagic aspirate which was inconclusive for diagnosis. Since investigations were not contributory an appropriate diagnosis could not be reached.

Due to financial constraints further investigations were not done. The lesion was excised through a transcervical approach. After raising the subplatysmal flaps, a bluish vascular lesion was noted in the floor of the submental

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triangle in the mylohyoid muscle (Fig. 1). The lesion was excised with a margin of surrounding normal muscle without significant bleeding (Fig. 2). Histological examination showed fragments of fibro adipose tissue showing a lesion composed of thick walled blood vessels between muscle fibers suggestive of cavernous IMH (Fig. 3). Patient is on regular follow up postoperatively for 1 year and no evidence of recurrence noted.

Fig. 1 Bluish vascular lesion in the submental region

Fig. 2 Specimen

Discussion

Liston in 1843 has described the first case and called this entity an ‘erectile tumor’. The etiology of this lesion is unknown; it is believed to develop from abnormal embryonal sequestrations. Muscular trauma has been proposed as an initiating factor and could account for why most occur in young population [4].

IMH is an uncommon, benign vascular neoplasm usually arising within the skeletal muscle of the trunk and the extremities. Though masseter has been the most common site in the head and neck region, IMH of the trapezius, orbital muscles, mentalis, digastric, geniohyoid, temporalis, sternocleidomastoid and mylohyoid have also been reported [5].

Fig. 3 Histopathological examination showing fragments of fibro adipose tissue with a lesion composed of thick walled blood vessels between muscle fibers suggestive of cavernous IMH, H&E stain, 20 × magnification

IMH usually presents as a distinct, localized, rubbery swelling in the second or third decades of life. Thrills, bruit, pulsation or skin discolorations usually associated with superficial haemangiomas are absent. Consistency may vary from soft and diffuse to firm and localized due to their deep location [3].

Diagnosing IMH can be difficult due to its rarity and non-specific clinical presentation. Plain radiography and FNAC are usually non-diagnostic. Sonography is usually the first line imaging procedure for patients with neck swellings but not specific for IMH. Color doppler sonography is useful to demonstrate the vascular structures in and around the muscle [6]. IMH often remains undiagnosed preoperatively, as in our case but Magnetic Resonance Imaging (MRI) is a reliable tool in detecting and delineating deep seated and large IMH [7]. Angiographic evaluations may detect feeding arteries in large IMH, but fail to be precise in small lesions. Nevertheless a pre-operative angiogram and embolisation can aid in reduction of perioperative haemorrhage [2, 4].

The histological appearance of the lesion is variable. These lesions are non-encapsulated benign neoplasms composed of differently sized vascular channels, sometimes with calcifications and admixed with variable amounts of adipose, fibrous and myxoid tissues. There may be evidence of inflammation and fibrous proliferation [4].

The differential diagnosis can be difficult and should include lymphadenopathy, salivary gland and muscular neoplasms, lymphangiomas and thyroid lesion.

Malignant transformation or spontaneous regression generally does not occur. Non-surgical methods like, steroid injection, embolisation, injection of sclerosing agents, cryotherapy, radiation therapy, laser therapy have been used in the past but are ineffective [8]. Complete wide excision with a cuff of the surrounding muscle is the

ideal treatment [3]. Recurrence in IMH is mainly due to incomplete excision.

Conclusion

Head and neck IMH is an uncommon, non-encapsulated vascular neoplasm characterized by painless and slow growth. IMH presents difficulties in correct preoperative diagnosis and treatment planning. Wide surgical resection beyond the gross limits of tumor is the most appropriate therapeutic option others being mostly palliative. The recurrence rate is high. Hence long term clinical and radiological follow up is recommended for timely diagnosis and treatment of recurrences.

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