CASE REPORT

Multidetector CT and histological features of benign mesenchymoma of the infratemporal space: a rare case report

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Benign mesenchymoma is a soft tissue neoplasm composed of an admixture of two or more benign mesenchymal components in addition to fibrous tissue. A rare case of benign mesenchymoma of the infratemporal space in a 14-year-old boy is presented. In this case report we discuss the salient imaging and histopathological features of this rare entity.

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Introduction

Mesenchymoma is a rare tumour of fibrous tissue origin composed of two or more mesenchymal derivatives.¹ The term mesenchymoma was first introduced by Klein¹ in 1932 to describe a primitive malignant mesenchymal neoplasm that showed marked histological similarity to Ewing's sarcoma. Despite its similarity to Ewing's sarcoma, all the elements constituting the tumour are well differentiated and do not show histological features of malignancy.² Typically, the tissues found in benign mesenchymoma include fat, smooth muscle and blood vessels, frequently with smooth muscle in their walls. Other tissue types including striated muscle, cartilage, bone, lymphoid and haematopoietic tissue have also been reported in these rare tumours. The proportions of the constituent elements vary widely from tumour to tumour so no two tumours are identical.

Most of these tumours are solitary and circumscribed but not encapsulated. They can infiltrate, if only microscopically, and their size can vary from 1 mm to 15 mm. Their growth rate is variable.³ In the head and neck region, benign mesenchymoma is a rare lesion.² Our review of the literature revealed 44 cases occurring

in the head and neck region. There has been no reported case of mesenchymoma occurring in the infratemporal space. In this case report, we describe an interesting case of a giant benign mesenchymoma in the infratemporal space and discuss the characteristic multi-detector CT (MDCT) imaging features of this condition.

Case report

A 14-year-old male presented with a history of a gradually progressive, painless, diffuse swelling on the right side of his face since early childhood (**Figure 1a**). Over the past few months, swelling had rapidly increased in size with significant cosmetic deformity. The patient had not experienced any difficulty in swallowing or chewing or ear ache. Clinical examination revealed a firm, immobile, non-fluctuant and non-tender mass in the temple (**Figure 1a**). Intraoral examination revealed a reddish swelling on the right buccal mucosa. Displacement of permanent maxillary right molars was observed without mobility (**Figure 1b**). There was no involvement of the facial nerve or the regional lymph nodes.

A MDCT examination was performed for further characterization of the swelling. Axial CT sections of

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b

Figure 1 (a) Clinical photograph showing enlargement of the right side of the face. (b) Intraoral photograph showing swelling of the right buccal mucosa and displacement of permanent maxillary right molars

a

b

Figure 2 Axial contrast enhanced CT images in the patient in (a) soft tissue window and (b) bone window demonstrating the mass in the right masticator space. (a) Axial contrast enhanced multi-detector CT images demonstrate the well encapsulated mass (thick arrows) in the right masticator space with fat component (thin arrow), bony component and soft tissue component. The parotid gland is displaced posterolaterally while the carotid vessels and jugular vein are not involved. (b) Axial CT image shows the osseous component of the lesion (black arrows). The mass is seen to cause anteromedial displacement and remodelling of the lateral wall of the maxillary sinus (thin arrow)

Figure 3 Axial multidetector CT image in the bone window demonstrates the bony component of the tumour (thick arrow) arising from the greater wing of temporal bone with adjacent sclerosis and remodelling of the temporal bone (thin arrows)

Figure 5 A photograph illustrating the transmandibular approach for removal of the tumour

the head and neck of 5 mm thickness were obtained (Figures 2 and 3). Coronal reformatted images of 3 mm thickness were obtained to demonstrate the extent of the tumour in the base of the skull. On MDCT, a large mass lesion was seen in the right masticator space extending into the infratemporal fossa with involvement of the right temporal bone. The tumour had a well-defined margin with heterogeneous density made up of soft tissue and fatty and bony components

Figure 4 Coronal reformatted multi-detector CT (MDCT) images demonstrating the extent of the tumour and the relationship to the skull base. (a) Coronal reformatted MDCT image in the soft tissue window demonstrates the extent of the tumour in the temporal fossa (thin arrow) with mass effect on the soft tissues of the oral cavity from the tumour. There is no destruction or involvement of the temporal lobe. (b) Coronal reformatted MDCT image in the bony window demonstrates absence of evidence of bony destruction of the skull base, supporting the benign nature of the tumour. There is remodelling and lateral displacement of the ramus of the mandible (black arrow)

Figure 6 Photograph showing multiple bits of the resected specimen. (Inset: cut surface showing yellowish-white areas probably representing adipose tissue)

(Figure 4). The mass caused distortion and displacement of the ramus of the mandible with anterolateral displacement of the ipsilateral zygomatic arch and anteromedial displacement of the lateral wall of the right maxillary sinus. There was no bony destruction seen. The extensive bony remodelling in the absence of

frank destruction was suggestive of a benign aetiology with features of slow growth. The mass caused posterior displacement of the ipsilateral carotid artery and the internal jugular chain without any evidence of infiltration.

Surgical resection of the tumour was performed extraorally using a transmandibular approach under general anaesthesia (Figure 5). The lesion appeared well demarcated and non-adherent to the surrounding tissues. No erosion of the adjacent bony surface was observed. Grossly the tumour consisted of multiple bits of firm, yellowish-white structures (Figure 6). The cut surface was yellowish-white in colour and gritty in consistency. The histopathological examination of the resected specimen revealed the presence of mature bone, cartilage and adipose tissue (Figure 7a,b). No glands or other well-formed epithelial structures were present, nor were cytological atypia or mitosis identified. A final diagnosis of a benign mesenchymoma of the infratemporal space was made.

Discussion

Benign mesenchymoma is an unusual neoplasm with unknown pathogenesis. Stout⁴ suggested that it may result from proliferation of pluripotent primitive

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Figure 7 (a) Photomicrograph showing mature bone, cartilage and fibrous tissue (haematoxylin and eosin stain, $\times 100$, black asterisk—cartilage, white asterisk—bone). (b) Photomicrograph showing adipose tissue (haematoxylin and eosin stain, $\times 100$)

Table 1 Details of benign mesenchymoma in the head and neck region

Reference	Age	Gender	Site	Diameter (mm)	Component tissues involved (other than fibrous tissue)										Prognosis
					Ad	Bv	Sm	St	Ly	My	Bo	Ca	Nf	Hi	
Le Ber and Stout ²	14 years	F	Eyelid	—	+	+	+								Dead ^a
Micheau et al ⁵	7 months	M	Tongue	5	+	+	+			+		+			NR (12 years)
Lavoie et al ⁶	9 years	F	Tongue	3	+	+	+	+			+				NR (2.5 years)
Cracovaner and Vieta ⁷	3 years	M	Tongue	5	+	+	+		+	+					NR (3 years)
Hatziotis et al ⁸	23 years	M	Tongue	1.8	+	+	+				+				U
Stanton ⁹	4 months	F	Neck (multiple)	1–1.5	+	+	+								U
Timar et al ¹⁰	5 weeks	F	Epiglottis	2.3	+	+	+			+					U
Janushka et al ¹¹	69 years	M	FOM	2.5	+	+	+								NR (14 years)
Janushka et al ¹¹	58 years	M	FOM	4	+	+	+								NR (8 months)
Bures and Barnes ¹²	14 years	F	Frontal scalp	2.5	+	+	+								NR (4 months)
Bures and Barnes ¹²	52 years	M	Cheek	1.7	+	+	+								NR (14 years)
Bures and Barnes ¹²	Newborn	M	Below ear	1	+	+	+		+	+					NR (13 years)
Bures and Barnes ¹²	19 years	M	Cheek	1.4	+	+	+								NR (6 years)
Bures and Barnes ¹²	Newborn	M	Trachea	1	+	+	+	+				+			Autopsy
Bures and Barnes ¹²	2 months	F	Frontal scalp	3.5	+	+	+	+							NR (5 months)
Bures and Barnes ¹²	7 months	F	Occipital scalp	2.2	+	+	+	+							U
Bures and Barnes ¹²	1 year	M	Neck	1.8	+	+	+								U
Nishijima et al ¹³	52 years	M	Palate	3	+	+	+			+		+			NR (16 months)
Freedman et al ¹⁴	2.5 years	F	Tongue	2.5	+	+	+								NR (10 months)
Makek and Lello ¹⁵	43 years	M	Palate	1.5	+	+	+						+		NR (21 months)
Makek and Lello ¹⁵	3 months	M	Tongue	2	+	+	+								NR (25 months)
Doraiswamy et al ¹⁶	Newborn	M	Neck	14	+	+	+					+			NR (21.5)
Couwenhoven et al ¹⁷	39 years	M	Lip	1.5	+	+	+	+							U
Dary and Guttenberg ¹⁸	55 years	F	Palate	1	+	+	+								NR (2 years)
Mohr et al ¹⁹	10 years	F	Tongue	4	+	+	+			+					—
Febro and Blas ²⁰	67 years	F	Hypopharynx	11	+	+	+								—
Darwin et al ²¹	78 years	F	Neck	3.5	+	+	+					+	+		U
Takeroshita et al ²²	88 years	F	Tongue	1.4	+	+	+			+					—
Robert and James ²³	20 years	F	Parapharynx	3	+	+	+					+			U
Fujiwara et al ²⁴	44 years	M	Cheek	3, 1.5	+	+	+		+	+	+		+		—
Sakurai et al ²⁵	6 years	M	Cheek	6	+	+	+							+	NR (3 years)
Yenchu et al ²⁶	32 years	M	Tongue	1.5	+	+	+			+	+	+			NR (6 months)
Jones et al ²⁷	60 years	M	Upper lip	0.75	+	+	+								—
Jones et al ²⁷	71 years	F	Buccal mucosa	4	+	+	+								—
Jones et al ²⁷	42 years	M	Buccal mucosa	3	+	+	+			+		+			—
Jones et al ²⁷	33 years	M	Lower lip	1.5	+	+	+								—
Jones et al ²⁷	55 years	M	Maxillary gingiva	1.5	+	+	+								—
Jones et al ²⁷	50 years	F	FOM	0.5	+	+	+								—
Jones et al ²⁷	56 years	M	Tongue	1	+	+	+					+			—
Jones et al ²⁷	40 years	M	Buccal mucosa	1.3	+	+	+					+	+		—
Jones et al ²⁷	66 years	M	Palate	0.2	+	+	+								—
Jones et al ²⁷	14 years	F	Tongue	1.5	+	+	+								—
Quinonez et al ²⁸	48 years	M	Tongue	5	+	+	+							+	—
Kessler et al ²⁹	60 years	M	Lower lip	1	+	+	+			+	+	+			—
Present case	14 years	M	Infratemporal space	4	+	+	+				+	+	+		—

Ad, adipose tissue; Bo, bone; Bv, blood vessels; Ca, cartilage; F, female; FOM, floor of mouth; Hi, histiocytic proliferation; Ly, lymphoid tissue, including lymphatic vessels; M, male; My, myxoid tissue; Nf, nerve fibres; NR, no recurrence [duration (in parentheses) indicates follow-up period]; Sm, smooth muscle; St, striated muscle; U, unknown.

^aDied during operation.

mesenchymal cells that undergo differentiation into various mesenchymal elements. The tumour is usually located in the extremities, kidney or urogenital tract, and its occurrence in the head and neck region is extremely rare.^{2,4} Our review of the literature found 44 cases of benign mesenchymoma reported in the head and neck region (Table 1).^{2,5–29} Although Le Ber and Stout² described a female predilection for this rare tumour, the majority of the mesenchymomas in the head and neck have been reported in males.^{2,5–29} Benign mesenchymoma in the head and neck do not have a particular age predilection; however, nearly 20.5% of them are detected in the first year of life. This tumour occurs predominantly in the intraoral location (60%), with the tongue being the most common site (26.7%). In the extraoral location (37.8%), they most commonly occur in the cheek.^{2,5–29} The infratemporal fossa is a very uncommon location for a benign mesenchymoma, and the tumour we encountered had an intraoral component in addition to an external swelling. In general, these tumours are well circumscribed but not encapsulated.² While the mesenchymoma is constituted by variable amounts of different histological components, adipose tissue is the most prominent component (82.2%) followed by vascular tissue (71%), smooth muscle (40%), cartilage (33.33%), bone (22.22%), myxoid tissue (22.22%), lymphoid tissue (13.33%), neural tissue (8.89%) and histiocytes (4.44%).^{2,5–29} It is still unclear whether this lesion represents a haematoma or a true neoplasm.¹²

The imaging appearance of this rare tumour reflects the histopathological heterogeneity. On MDCT, mesenchymoma has a heterogeneous appearance composed of fatty, soft tissue density and bony components. Cross-sectional imaging, particularly MDCT, plays a crucial role in dictating treatment strategies, including surgical approach by defining the tumour planes and

extent of involvement. MDCT chiefly helps in defining the tumour margin and the relationship of the tumour to the vital structures in the skull base, including the carotid vessels, internal jugular chain and cranial nerves (trigeminal nerve and facial nerve). Multiplanar coronal and sagittal MDCT reformations are very valuable for pre-operative surgical planning as the three-dimensional relationship of the tumour to the vital structures in the skull base is clearly illustrated. They offer valuable pre-operative delineation of these anatomical relationships which is imperative to minimize post-operative damage to nearby vessels and nerves and limit post-operative morbidity. The differential diagnosis for these fat- and bone-containing tumours in the neck includes teratoma, ossifying lipoma and liposarcoma. The absence of bone destruction and infiltration to surrounding vessels is an important sign to rule out a malignant neoplasm. As seen in our case, the presence of bone remodelling also gives vital clues to the benign nature and slow growth of the tumour. Although MRI was not performed in our patient, it would provide additional information regarding the relationship of the tumour to the cranial nerves in the skull base.

Complete surgical excision is the treatment of choice for most benign mesenchymomas. Roughly 20% of benign mesenchymomas recur after surgical excision in other parts of body; however, there are no reported cases of recurrent tumour in the head and neck.² A long-term follow-up is justified for tumours in the head and neck owing to the high rate of recurrence elsewhere in the body; however, close imaging surveillance may not be mandated.

To summarise, benign mesenchymoma is a rare tumour of mesenchymal origin. When encountered in the head and neck, a detailed evaluation with cross-sectional imaging studies like MDCT is necessary for treatment planning and for determining surgical approach.

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