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Masson's Tumour of Parotid: A Case Report and Literature Review

Parotis Bezinin Masson Tümörü: Olgu Sunumu ve Literatür Derlemesi

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ABSTRACT

Masson's tumour is a rare non-neoplastic vascular lesion characterized by a reactive proliferation of endothelial cells with papillary growth secondary to intravascular thrombosis or vascular stasis. It most commonly affects medium-sized veins and mainly involves the skin and subcutaneous tissues of the head and neck region, trunk, and extremities. Parotid gland involvement is extremely rare, with only four cases reported worldwide to date. The preoperative diagnosis is challenging, and the condition is usually misdiagnosed as another more common benign lesion. Histopathological examination and immunohistochemical studies are considered the gold standard for the diagnosis and exclusion of malignant lesions that may resemble Masson's tumour. The extent of surgical resection, either superficial or total parotidectomy, depends on preoperative imaging and intraoperative findings. The prognosis is favourable with a low recurrence rate if the surgical margin is negative.

Keywords: Masson's tumour, intravascular papillary endothelial hyperplasia, parotid gland, parotidectomy

Öz

Masson tümörü, intravasküler tromboz veya vasküler staza sekonder olarak papiller büyüme gösteren reaktif endotelial hücre proliferasyonu ile karakterize, nadir görülen ve neoplastik olmayan bir vasküler lezyondur. En sık orta büyüklükteki venleri etkiler ve baş-boyun bölgesi, gövde ve ekstremitelerin deri ile subkutan dokularını tutar. Parotis bezi tutulumu son derece nadirdir ve günümüze kadar dünya çapında yalnızca dört vaka bildirilmiştir. Preoperatif tanı güç olup, genellikle daha yaygın görülen başka bir benign lezyon olarak yanlış değerlendirilir. Histopatolojik inceleme ve immünohistokimyasal çalışmalar, Masson tümörüne benzer malign lezyonların ayırıcı tanısı ve doğrulanmasında altın standarttır. Cerrahi rezeksiyonun kapsamı, yüzeysel veya total parotidektomi olmasına dayalı olarak preoperatif görüntüleme ve intraoperatif bulgulara bağlıdır. Cerrahi sınır negatif olduğunda prognoz olumludur ve nüks oranı düşüktür.

Anahtar Sözcükler: Masson tümörü, intravasküler papiller endotelial hiperplazi, parotis bezi, parotidektomi

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INTRODUCTION

Masson's tumour, also known as intravascular papillary endothelial hyperplasia (IPEH), is a rare, non-neoplastic lesion of vascular origin, characterized by reactive proliferation of endothelial cells with papillary growth (1,2). Pierre Masson, a French pathologist, first described this disease in 1923 as "vegetant intravascular hemangioendothelioma" (3).

The most common sites of involvement are the skin and subcutis and comprises around 2% of vascular tumours of the skin and subcutaneous tissue (4-6). It can occur in any blood vessel but has a predilection for the skin and subcutaneous tissues of the head and neck region, trunk, and extremities (5-7). The skin and subcutaneous tissue of the face and scalp are the most commonly affected sites in the head and neck region (7). Parotid gland involvement is extremely rare, with only four cases reported worldwide to date (5,6,8,9).

CASE REPORT

A 50-year-old male with underlying diabetes mellitus and hypertension presented with a one-year history of painless right parotid swelling that had gradually increased in size. There were no other locoregional symptoms, such as facial weakness, trismus, dysphagia, voice changes, or neck swelling. He has no risk factors for malignancy, such as smoking, alcohol consumption, a family history of malignancy, or a history of head and neck radiation. In addition, he denied any constitutional symptoms.

On examination, there was a diffuse swelling in the right parotid region, which had a smooth surface, was firm in consistency, non-tender, and immobile but not fixed to the overlying skin (Figure 1). The facial nerve was intact, with no medialisation of the lateral pharyngeal wall and no palpable cervical lymph nodes. Other head and neck examinations were unremarkable.

A computed tomography scan of the neck was performed and showed a well-circumscribed, heterogeneously enhancing solid lesion within the deep lobe of the right parotid gland, measuring $2.7 \times 3.2 \times 3.9$ cm (Figure 2). There was no calcification, bony erosion, or cervical lymph node enlargement, and the superficial lobe appeared normal. A through-cut biopsy was performed, which



Figure 1. Diffused swelling at the right parotid region.

revealed normal salivary gland tissue. This finding likely represented the normal superficial lobe of the parotid gland rather than the deep lobe lesion.

The patient subsequently underwent a total parotidectomy. Intraoperatively, the mass was confirmed to arise from the deep lobe of the parotid gland, whereas the superficial lobe appeared normal (Figure 3). Postoperatively, the patient recovered well without any complications. Histopathological examination (HPE) of the deep lobe revealed an intrasalivary gland lesion, surrounded by a pseudofibrous capsule and composed of variably sized vascular spaces, with some areas exhibiting papillary structures within (Figure 4A). The papillae are lined by a single layer of plump endothelial cells (Figure 4B). No nuclear atypia or mitotic figures seen. Most areas exhibit organised

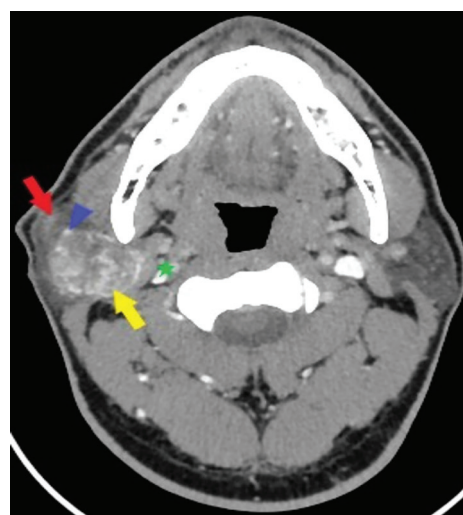


Figure 2. Contrasted axial view computerized tomography scan shows a well circumscribed heterogeneously enhancing solid lesion seen within the deep lobe of the right parotid gland (yellow arrow), without extension into the parapharyngeal space (green star). The lateral lobe appeared normal (red arrow). Blue arrowhead – retromandibular vein.

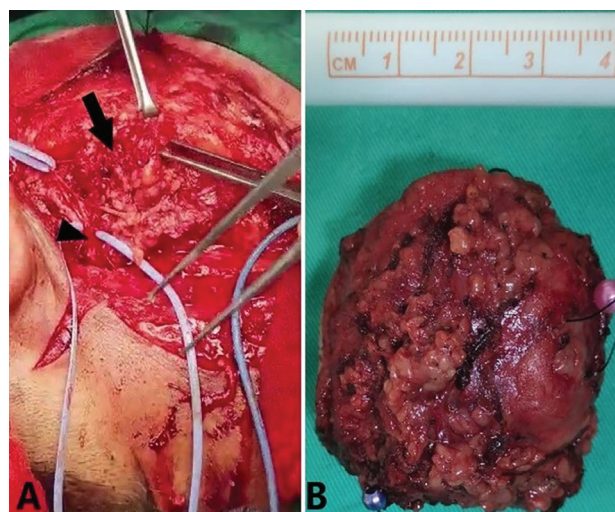


Figure 3. Mass arises from the deep lobe of parotid gland (arrow), deeper to facial nerve (arrowhead – main trunk of facial nerve) (A). A 4cm x 4cm mass with deep lobe of parotid gland was completely excised (B).

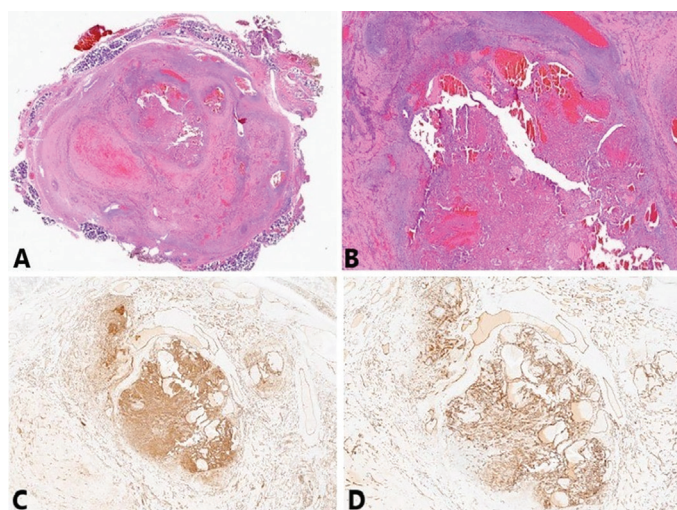


Figure 4. An intra-salivary gland lesion, surrounded by pseudo-fibrous capsule, composed of variable sizes of vascular spaces, with areas of papillary structures within that are lined by single layer endothelial cells (A, H&E x 40). Focal areas with papillary structures lined by endothelial cells (B, H&E x 400). CD31 immunostaining (C, x 200) and CD34 immunostaining (D, x 200) highlighting the endothelial cells lining the vessel wall.

thrombi associated with stromal haemorrhage, hyalinisation, myxoid change, and aggregates of haemosiderin-laden pigments. The endothelial cells lining the vascular channels are immunoreactive for CD31 and CD34 (Figure 4C and 4D) and negative for CK7, CK20, p63, and HHV8. These findings are suggestive of Masson's tumour. The HPE of the superficial lobe was normal.

DISCUSSION

Masson's tumour is a rare vascular lesion that mostly affects medium-sized veins but may occur in veins of any size and, rarely, in arteries (5,9). It was first described by Pierre Masson in 1923, who used the term vegetant intravascular hemangioendothelioma when he discovered intravascular papillary proliferation in a male patient with an inflamed hemorrhoidal plexus (3). It is also known as IPEH, a term highlighted by Clearkin and Enzinger (10) in 1976 to emphasize the non-neoplastic nature of the disease. Other terms used in the past to describe this disease entity include intravascular angiomatosis, intravenous atypical vascular proliferation, intravascular endothelial proliferation, reactive papillary endothelial hyperplasia, Masson's pseudoangiosarcoma, and Masson's intravascular hemangioendothelioma (1,6,7).

Generally, the skin and subcutis are the most commonly affected sites (4-6), while the face and scalp are the most frequently reported sites in the head and neck region (7). Oral cavity, lateral neck, mandible, thyroid, orbit, internal auditory canal, pharynx, larynx, maxillary sinus, and intracranium are among the reported sites of involvement in the head and neck region (1,6,7). Based on a literature review, salivary gland involvement is extremely rare, with only four cases reported in the parotid gland and one case in the submandibular gland (Table 1) (1,5,6,8,9). A case reported by Corio et al. (11) in 1982 was excluded due to the absence of clinical details, and another case reported by Kim et al. (12) in 2007 was also excluded because the mass was separate and located outside the submandibular gland.

Masson's tumour commonly occurred in adults without a distinct age, sex, or racial predilection (2,4). Incidence in children is very low, but it can occur as early as 9 months of age (7). Some studies suggest a slight female predominance, with a female-to-male ratio of 1.3:1.0 (1,6). It is a generally slow-growing tumour and usually presents

Table 1. Review of reported cases of salivary gland Masson's tumour

Author	Year	Age	Sex	Location	Presentation	Imaging	Biopsy/FNAC	Clinical impression	Treatment
Mokhtari et al. (8)	2011	39-year-old	M	Left parotid gland	Painless swelling for 1 year	NA	NA	Pleomorphic adenoma	Partial resection of the parotid gland
Mignogna et al. (6)	2016	70-year-old	F	Right parotid gland	Painless swelling for 5 years	MRI	NA	Pleomorphic adenoma	Extracapsular dissection
Carta et al. (9)	2018	43-year-old	F	Right parotid gland (superficial lobe)	Mild painful swelling for 4 years	MRI	NA	A hyper vascularized benign lesion	Subtotal parotidectomy
Cho et al. (1)	2021	37-year-old	F	Right submandibular gland	Painless swelling for 2 months	US and CT scan	Presence of some fibrinous material and mixed inflammatory cells	Sialadenitis secondary to sialolithiasis with intraglandular abscess or submandibular gland tumour	Ablation of the submandibular gland
Mohebbi et al. (5)	2023	29-year-old	F	Right parotid gland (superficial lobe)	Painless swelling (duration not mentioned)	MRI	NA	A vascular mass	Total parotidectomy

M: Male, F: Female, US: Ultrasound, CT: Computerised tomography, FNAC: Fine needle aspiration cytology, MRI: Magnetic resonance imaging, NA: Not available

with painless swelling (7). The aetiology and pathogenesis are poorly understood, with various theories having been hypothesised. The aetiology is believed to be multifactorial, with approximately 30% of cases triggered by local trauma and vascular anomalies (2,4,5). Intravascular thrombus formation is the key factor in several theories of pathogenesis. Masson first described the disease as a neoplasm characterised by endothelial proliferation which results in thrombosis (3). Fifty-three years later, Clearkin and Enzinger (10) proposed a more widely accepted theory that explained that the formation of thrombosis leads to reactive proliferation of endothelial cells and papillary formation. This finding emphasized that the lesion is a reactive process rather than a neoplasm. Another theory by Levere et al. (13) is that the release of basic fibroblast growth factor by macrophages in response to traumatic vascular stasis stimulates endothelial cell proliferation. Additionally, Irey and Noris (14) suggested that hormones may play a role in the pathogenesis due to the female predilection in some studies.

Hashimoto et al. (15) classified Masson's tumour into 3 types based on aetiology. The pure type is the most common and arises de novo in a dilated vessel; the mixed type is associated with a pre-existing vascular anomaly; the extravascular type is associated with trauma-induced hematoma formation. Preoperative diagnosis is challenging. Fine-needle aspiration cytology is usually non-diagnostic or results in misdiagnosis in the majority of cases (7). Our present case underwent a Tru-Cut biopsy; however, the sample was not representative of the lesion. Imaging studies, whether computed tomography or magnetic resonance imaging, are non-specific but may be helpful in surgical planning (16). In the parotid gland, imaging features may resemble benign lesions such as pleomorphic adenoma, Warthin's tumour, or vascular tumours.

The diagnosis of Masson's tumour is usually made after HPE and immunohistochemistry (IHC) studies of the excised specimen. They are considered the gold standard for the diagnosis of Masson's tumour and the exclusion of malignant lesions that may resemble Masson's tumour, such as angiosarcoma and Kaposi sarcoma. The typical histopathological findings of Masson's tumour are proliferation of papillary structures within the blood vessel, thrombus formation, coverage by fewer than three layers of endothelial cells, and positive CD31 and CD34 immunohistochemical markers (1). On the other hand, proliferation of cells outside the vessel, papillae covered by more than two endothelial cell layers, and a positive CD105 immunohistochemical marker are more suggestive of angiosarcoma (1).

The treatment of Masson's tumour is complete excision with negative margins. The extent of surgical resection depends on the evaluation of preoperative imaging and intraoperative findings. A superficial parotidectomy is sufficient if the lesion is confined to the superficial lobe of the parotid without features suggestive of malignancy. However, if the lesion is located in the deep lobe of the parotid gland, total parotidectomy is usually required, as observed in the present case. The prognosis is good, with a low recurrence rate, if the margin is negative (1).

CONCLUSION

Masson's tumour is extremely rare occurred in parotid gland and usually misdiagnosed as another more common benign lesion.

Preoperative diagnosis is challenging, and most cases are diagnosed after HPE and IHC studies of the excised specimen. It is important to exclude other more serious conditions that may resemble Masson's tumour, such as angiosarcoma and Kaposi sarcoma. The treatment consists of complete excision with negative margins, and the prognosis is generally good, with a low recurrence rate.

Ethics

Informed Consent: Informed consent was obtained for publication.

Footnotes

Authorship Contributions

Surgical and Medical Practices: V.S.K.E.D., O.W.K., L.S.L., Concept: V.S.K.E.D., L.S.L., Design: O.W.K., L.S.L., Data Collection or Processing: V.S.K.E.D., O.W.K., L.S.L., Analysis or Interpretation: V.S.K.E.D., O.W.K., L.S.L., Literature Search: V.S.K.E.D., O.W.K., Writing: V.S.K.E.D.

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