

The Grody Turf-Osseous Choristoma

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Preface

Choristoma is defined as a tumour-like proliferation of normal tissue which appears localized within an ectopic or anomalous location. Initially scripted by Albrecht in 1904, choristoma exemplifies a "tumour-like lesion" which is essentially comprised of normal tissue confined to an aberrant region [1]. Subsequently, Monserrat in 1913 defined a bone-forming neoplasm of the tongue designated as "lingual osteoma" [2]. Choristoma is composed of proliferating, histologically normal tissue which accumulates within an aberrant anatomic site. Thus, choristoma is essentially denominated by a tumour-like mass constituted of normal cells confined to an anomalous location. Choristoma of the oral cavity is infrequent and preponderantly composed of bone or cartilage. Nevertheless, choristoma may be comprised of peripheral glial tissue, sebaceous glands or gastrointestinal tissue. Choristoma is generally devoid of salivary gland ductal tissue or a myoepithelial component.

Disease Characteristics

Choristoma may arise from diverse tissues as the bone, cartilage, lingual thyroid, salivary gland, glial tissue, gastric mucosa or accessory soft tissue [3,4]. Oral osseous choristoma is frequently confined to dorsum of the tongue and usually appears adjacent to foramen caecum and circumvallate papillae, lateral border or the left, right or midline of posterior region of the tongue. Majority (~85%) of osseous and cartilaginous choristoma are situated upon the tongue, especially posterior third of dorsum of tongue, abutting the foramen cecum [3,4]. Majority of instances appear between second decade to third decade although the lesion may be discerned between first decade to eighth decade and no age of disease occurrence is exempt [3,4]. Exceeding > 70% of lingual, osseous and cartilaginous choristomas are manifest within the female population. A female predominance is observed with a female to male

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proportion of ~ 2.5 :1. Lesions may be delineated within 3 days to 50 years [3,4]. Appropriate categorization of oral choristoma is contingent to subtype of tissues constituting the choristoma [3,4]. Salivary gland choristoma is divided into central choristoma or gingival choristoma, lesions which demonstrate ectopic salivary gland tissue. However, the raised, tumour-like mass is devoid of contiguity with normal major or minor salivary glands [3,4]. Cartilaginous choristoma is comprised of multiple lobules of mature hyaline cartilage embedded within perichondrium-like fibrous tissue which resembles perichondrium. Constituting chondrocytes may be miniature to enlarged although cellular atypia is absent [3,4]. Additionally, variants such as lingual thyroid choristoma, lingual sebaceous choristoma or glial choristoma may be observed. Choristoma composed of gastric or respiratory mucosa may be exemplified as solid or cystic lesions [3,4].

Disease Pathogenesis

Of obscure aetiology and pathogenesis, osseous choristoma may emerge as a developmental disorder and is posited to arise as a posttraumatic or reactive lesion [3,4]. Additionally, osseous choristoma may occur due to congenital malformation, preceding trauma or chronic lingual irritation [4,5]. Although of undetermined aetiology and pathogenesis, osseous choristoma is postulated to be a malformation or may arise due to trauma or chronic irritation [4,5].

• Malformation hypothesis surmises that the lesion arises upon line of fusion between first and third branchial arches, particularly upon anterior two thirds and posterior one third of the tongue. It is documented that normal osseous structures as the incus, malleus and hyoid bone are derived from first and third branchial arches respectively. Entrapped, pluripotent cells generated from aforesaid branchial arches subsequently undergo ossification with the emergence of osseous choristoma [4,5]. • Trauma hypothesis indicates the predilection for posterior third of tongue towards trauma and irritation with consequent occurrence of an osseous lingual lesion with a reactive or post-traumatic centric ossification [4,5].

However, normal bone formation may ensue within osseous choristoma rather than the irregular bone devoid of Haversian canal system occurring with traumatic ossification [4,5].

Clinical Elucidation

Clinical symptoms may be contingent to magnitude and location of lesion. Lingual osseous choristoma emerges as a painless, firm, exophytic, pedunculated nodule with a narrow base. The lesion is usually confined to posterior, midline or lateral margin of the tongue [5,6]. Oral osseous choristoma may manifest as an asymptomatic, self-limiting lesion or a lingual mass which engenders nausea, discomfort, foreign body sensation or a gagging sensation, mass effect, hoarseness, swelling, pain, excessive cough, dysphonia, reflexive vomiting or dysphagia. Additionally, tongue discharge, respiratory disturbances, irritation or restricted tongue movements may ensue [5,6].

Histological Elucidation

Upon gross examination, the spherical, sessile or pedunculated, well circumscribed, firm to hard, dense tumefaction with a smooth outline and magnitude varying from 0.3 centimetres to 5 centimeters and a superimposed intact, normal mucosa may be observed. Nevertheless, superficial mucosa may be ulcerated. Pedunculated masses may exhibit a narrow base [6,7]. Upon microscopy, the mass is comprised of circumscribed mature, lamellar bone with enveloping benign, stratified squamous epithelium. Fragments of mature cartilage admixed with mature, compact bone constituted of osteocytes or osteoblasts along with an intact Haversian canal system may be observed. The tumefaction is superimposed by an unremarkable stratified squamous epithelium [6,7].

Osseous choristoma is composed of dense, mature bone wherein normal osteocytes are compact and osteoblastic rimming is absent. Occasionally, bone and cartilage may concur within a singular lesion [6,7].



Figure 1: Osseous choristoma depicting a pedunculated, non-erythematous growth with a smooth, non-ulcerated surface and a narrow base [8].



Figure 2: Osseous choristoma exhibiting foci of mature bone imbued with osteocytes and a normal Haversian system [9].

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Figure 3: Osseous choristoma exemplifying foci of mature cartilage and bone admixed with a fibrous tissue stroma and a superimposed unremarkable stratified squamous epithelium [10].



Figure 4: Osseous choristoma depicting mature bone with an intact Haversian canal system and commingled fibrous tissue [11].



Figure 5: Osseous choristoma enunciating foci of mature bone, cartilaginous tissue, patent Haversian canals and intermingled fibrous tissue [12].



Figure 6: Osseous choristoma displaying Haversian canals with osteocytes, mature bone and circumscribing fibrous tissue stroma [12].

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Figure 7: Osseous choristoma demonstrating a lace-like pattern of mature bone, admixed fibrous tissue and a superimposed, unremarkable fibrous tissue stroma [13].



Figure 8: Osseous choristoma exhibiting a nodular, sessile, bony mass composed of mature bone and superimposed stratified squamous epithelium [14].

Differential Diagnosis

Clinical segregation of osseous choristoma from tumour-like lesions, benign neoplasms and malignant conditions such as pyogenic granuloma, haemangioma or squamous cell carcinoma is necessitated [7,15]. Protrusive tumefaction confined to dorsum of the tongue, abutting the foramen caecum requires a distinction from benign neoplasms as a haemangioma, lymphangioma, teratoma, hamartoma, leiomyoma, thyroglossal duct cyst, lingual thyroid, mucocoele or pyogenic granuloma and malignant neoplasms as rhabdomyosarcoma or epidermoid carcinoma [7,15]. Peripheral lingual lesions require a demarcation from conditions such as traumatic neuroma, neurofibroma, schwannoma, fibroma or cartilaginous choristoma [7,15]. Anterior lingual region commonly delineates nodules of pyogenic granuloma, mucocoele or cartilaginous choristoma [7,15].

Hamartoma is constituted of disorganized proliferation

of mature tissues which are normally configured at specific sites. Emergence of a hamartoma is associated with a singular, predominating tissue element. Therefore, hamartoma is described as a tumour-like malformation or a disorganized mass comprising of mature, normal tissue or visceral cells situated within the usual location [7,15].

Cartilaginous metaplasia is usually confined to soft tissues situated beneath improperly fitting dentures. The lesion may depict diffusely disseminated calcium deposits and scattered, singular or clustered foci of cartilaginous cells configuring diverse stages of cellular maturation [7,15].

Pleomorphic adenoma of the salivary gland may demonstrate foci of osteo-cartilaginous tissue. Typically, the neoplasm exhibits cysts and tubules coated with an epithelial (ductal) component configuring intrinsic layer and an admixture of myoepithelial cells disseminated within the extrinsic layer. Myoepithelial cells may appear scattered within the myxoid stroma and represent as

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plasmacytoid, spindle-shaped, epithelioid, stellate- shaped or clear cells. Circumscribing stroma is myxoid, chondroid, myxochondroid, hyalinised or fibrotic [7,15]. Additionally, segregation is necessitated from vascular malformations, fibroma, papilloma, pleomorphic adenoma of the salivary gland, osteochondroma and malignant neoplasms as osteogenic sarcoma or squamous cell carcinoma [7,15].

Investigative Assay

Lesions exceeding >2 centimetre magnitude necessitate appropriate tissue sampling. Upon computerized tomography (CT), lingual osseous choristoma manifests as a well-defined, spheroid, bone-attenuating tumefaction which is commonly situated upon base of the tongue. Continuity of the tumefaction with circumscribing soft tissues can be evaluated [16,17]. Magnetic resonance imaging (MRI) may also be adopted to assess the neoplasm [16,17].

Therapeutic Options

Optimal treatment of lingual osseous choristoma is comprehensive surgical extermination of the lesion. Alternatively, carbon dioxide laser or potassium titanyl phosphate (KTP) laser therapy or electrosurgical eradication of the lesion may be beneficially adopted [16,17]. Occasionally, lingual osseous choristoma with miniature pedicle may be eradicated with coughing or swallowing. Thus, a conservative approach may be recommended for treating an asymptomatic, miniature, pedunculated, lingual osseous choristoma which may resolve spontaneously [16,17]. Prognostic outcome of surgically resected osseous choristoma is favourable. Tumour reoccurrence is minimal and may be observed in osseous choristoma of the masseter [16,17]. However, non-lingual osseous choristoma arising within gingiva or cheek may exhibit reoccurrence and can be adequately determined by repetitive histological examination. Malignant metamorphosis is usually absent [16,17].

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