Pattern of presentation and management of orofacial and neck soft tissue tumors in a Nigerian tertiary health center - a preliminary study

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Abstract

Background: The purpose of this study was to document the pattern of presentation and management of orofacial and neck soft tissue tumors seen in a Nigerian tertiary health center.

Method: Lesions included were hamartomas, benign and malignant tumors. The extent of spread based on the involvement of either the skin, subcutaneous, mucosa alone or in combination was noted. Involvement was determined based on clinical assessment. Histopathological examination of the lesions was reviewed. Treatment offered and outcome was also documented.

Result: A total of 60 patients with tumors of the orofacial and neck soft tissues were managed in our hospital. Males constituted 33 (54.6%) and females were 27 (45.4%). Age range was 6 months to 66 years and mean age (SD) was 35.4 (15.1) years. Surgical excision was done for all hamartomas and benign tumors.

Conclusion: Aesthetic and functional outcome was satisfactory in all the patients except for a case of malignant schwannoma with multiple recurrences.

Keywords: Pattern, management, soft tissue, orofacial, neck, tumors

Introduction

The overlying soft tissues of the head and neck comprise mainly of subcutaneous fat, neurovascular tissues, fascia and muscles covered by skin and lined by mucosa. Parenchyma organs, glands, glia and lymphoreticular tissues are not classified as soft tissues [1]. Tumors arise from any part of the soft tissues and spread to involve the mucodermal tissues to different extent. Rate of growth and extent of involvement depend on the nature of the lesion. The categories of lesions of soft tissues include hamartomas, benign and malignant tumours [2]. The treatment and outcome of these lesions are determined by the extent of spread/stage of the disease [3]. When lesion is confined within the subcutaneous plane, there might be no need to utilize flaps for repair especially when collapse will not be appreciable, however when significant mucodermal tissue is lost, aesthetics becomes a challenge to overcome. Our department is a relatively new center with a low turn up of these cases and there has been scanty report of these lesions from other parts of the country. The aim of this study was therefore to document the pattern of presentation and management of orofacial and neck soft tissue tumors seen in our center in the last 6 years.

Patients and methods

This was a retrospective study that included consecutive patients present to the Oral and maxillofacial clinic of the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria with

orofacial and neck soft tissue tumors between May 2006 and August 2012. Salivary glands, head/neck viscera and lymph node tumors were excluded. Patients' demographics and clinical presentation of the tumors were retrieved from the case files, theater registers and histopathology department electronic database. Lesions included were hamartomas, benign and malignant tumors. The extent of local spread based on the involvement of either the skin, subcutaneous, mucosa alone or in combination was noted. Involvement was determined based on the presence of discoloration, erythema, maculopapular or nodular elevations, erosion, atrophy, ulceration, fungation or necrosis of any varying sizes and extent. Local, regional and distant lymph node enlargements were also documented. Chest X-rays, plain radiograph skeletal surveys and ultrasound investigations that were done were reported. Histopathological examination of the lesions was reviewed. Treatment offered (excision, neck dissection, and primary closure, reconstruction with flaps, radiotherapy, and chemotherapy), outcome and follow up reviews were also documented. The study was performed according to the ethical guidelines for research involving human subjects and ethical clearance obtained from the hospital's ethics committee. Data obtained was documented, analyzed and expressed as simple frequencies and proportions with SPSS package version 16.

Result

A total of 60 patients with tumors of the orofacial and neck

Table 1. Age and site distribution of patients with orofacial and neck soft tissue tumors.

Age	Benign/Hamartoma		Malignant		Total N(%)	Site	N(%)
	M(%)	F(%)	M(%)	F(%)			
0-9yrs	1(1.7)	4(6.8)	0(0)	0(0)	5(8.5)	Scalp	19(31.7)
10-19yrs	2(3.4)	0(0)	0(0)	1(1.7)	3(5.1)	Eyelid	1(1.7)
20-29yrs	3(5.0)	5(8.5)	0(0)	1(1.7)	9(15.0)	Cheek	10(16.7)
30-39yrs	12(20.0)	6(10.0)	1(1.7)	1(1.7)	20(33.3)	Lip	5(8.4)
40-49yrs	4(6.8)	4(6.8)	1(1.7)	0(0)	9(15.0)	Palate	1(1.7)
50-59yrs	7(11.9)	4(6.8)	1(1.7)	1(1.7)	13(21.8)	Tongue	2(3.4)
60-69yrs	0(0)	0(0)	1(1.7)	0(0)	1(1.7)	Neck	10(13.3)
	--	--	--	--	--	Multiple	6(10)
Total	29(47.8)	23(38.6)	4(6.8)	4(6.8)	60(100)	Total	60(100)

Table 2. Gender distribution of Hamartomas, benign tumours and malignant tumours.

Hamartomas/Benign	Male N(%)	Female N(%)	N(%)
Hemangioma	4(6.7)	4(6.7)	8(13.3)
Neurofibroma	3(5.0)	3(5.0)	6(10.0)
Lipoma	18(30.0)	11(18.3)	29(48.3)
Papilloma	2(3.3)	0(0)	2(3.3)
Rhabdomyoma	1(1.7)	0(0)	1(1.7)
Dermatofibroma	3(5.0)	3(5.0)	6(10.0)
Malignant			
Rhabdomyosarcoma	1(1.7)	1(1.7)	2(3.4)
Mucoepidermoid carcinoma	1(1.7)	0(0)	1(1.7)
Malignant Schwannoma	1(1.7)	0(0)	1(1.7)
Kaposi Sarcoma	1(1.7)	0(0)	1(1.7)
Squamous cell carcinoma	1(1.7)	1(1.7)	2(3.4)
Fibrosarcoma	0(0)	1(1.7)	1(1.7)
Total	36(60.2)	24(38.8)	60(100)

soft tissues were managed in our hospital. Males constituted 33(54.6%) and females were 27(45.4%). Age range was 6 months to 66 years and mean age (SD), was 35.4(15.1) years. There were more patients with hamartoma/benign tumors 52(86.4%) than malignant tumors 8(13.6%). One-third of the patients (33.3%) were in fourth decade of life followed by 13(21.8%) in the sixth decade (**Table 1**).

Hamartomas constituted 14(23.3% out of which hemangioma was 8(13.3%) and neurofibroma 6(10%). Lipoma was the commonest of the benign tumor and all the lesions, constituting 29(48.3%) followed by dermatofibroma 6(10%) (**Table 2**).

Most of the tumors were found on the scalp 19(31.7%) closely followed by the cheek 10(16.7%), neck 8(13.3%) and lip 5(8.3%) (**Table 1**). Six (10%) patients had lesions in multiple sites which include scalp, face, neck and trunk. These lesions in multiple sites were neurofibroma, hemangioma (**Figure 1**)

Figure 1. Photomicrograph of carvenous heamangioma showing large endothelial lined vascular channels containing red blood cells (arrow). H&E x 10.

and lipoma (**Figure 2**).

Malignant lesions that involve the tongue affected the mucosa, submucosa and muscles and these are well differentiated squamous cell carcinoma of granular type with prominent keratin pearls and distinct squamous epithelial cells (3.4%) (**Figure 3**) and mucoepidermoid carcinoma with darkly stained epidermoid cells and clear mucoid cells 1(1.7%) (**Figure 4**). Four (6.7%) malignant lesions at presentation had spread locally to involve the skin, subcutaneous tissue and mucosa. These are pleomorphic rhabdomyosarcoma (**Figure 5**) fibrosarcoma (**Figure 6**) and malignant schwannoma (**Figure 7a** and **7b**), all affecting the cheek and lip, while Kaposi sarcoma of the palate was limited to the mucosa (**Figure 8**). Lipoma 29(100%) involved mainly the subcutaneous tissue and buccal pad of fat, also hemangioma affected the three layers of tissues especially when found in the region of the cheeks and lips.

Plain X-rays did not show evidence of local or distant metastases. Ultrasound was done in 5% of cases of the lesions; reports were suggestive but not conclusive in those

Figure 2. Photomicrograph of lipoma showing mature fat cells (adipocytes) with peripherally displaced nuclei (arrow). H&E x10.

Figure 4. Photomicrograph of mucoepidermoid carcinoma showing part of the capsule on the upper part and darkly stained epidermoid cells and clear mucoid cells within the lesion (arrow). H&E x10.

Figure 3. Photomicrograph of squamous cell carcinoma showing islands of neoplastic well differentiated squamous cells within the connective tissue. Also seen with the tumour islands are keratin pearls (arrow). H&E x10.

Figure 5. Photomicrograph of rhabdomyosarcoma (pleomorphic type) showing malignant round and spindle muscle cells mitotic figures and tadpole's cell (arrow).

cases. Lipoma and hemangioma showed varying degrees of echodensities but hemangioma was poorly delineated compared to lipoma which showed higher echodensities and better delineation.

All the lesions were treated by surgical excision out of which 58(96.6%) was accompanied with primary closure, that on the palate healed by secondary intention and the patient with malignant schwannoma had closure with regional cervical flap for mucosal lining and delto-pectoral flap for covering.

The distant flap was also used for reconstruction of the lower lip during inset.

The cases of squamous cell carcinoma of the tongue involved less than one quarter of the tongue, closure was achieved with primary closure, the speech and swallowing functions were not significantly affected by the tissue loss. All the cases of malignant epithelial lesions were also referred for radiotherapy after excision of the lesion at the primary site. Type 3 functional neck dissection was done in one of the cases of squamous cell carcinoma based on clinical diagnosis of enlarged cervical node. Patients with rhabdomyosarcoma

Figure 6. Photomicrograph of fibrosarcoma showing neoplastic plump and angular fibroblasts in herring bone pattern (arrow).

Figure 8. Photomicrograph Kaposi sarcoma demonstrating the proliferation of endothelial cells and numerous slit-like vascular spaces (hematoxylin and eosin, magnification $\times 200$).

Figure 7. (a) Photomicrograph of malignant nerve sheath tumour showing malignant nerve cells in a wavy pattern. H&E $\times 10$. (b) High power view of the malignant nerve cells showing evidence of pleomorphism. H&E $\times 40$.

were treated by combination of surgery and chemotherapy. There was no case of recurrence in the hamartoma and benign tumors while 1(1.7%) case of multiple recurrences was reported in the patient with malignant schwannoma.

Discussion

The prevalence of soft tissue tumors in the head and neck region in our hospital was quite low and this could possibly be attributed to multiple health care facilities in our region, inability of low socioeconomic status patients to afford hospital care and an actual low occurrence of these lesions. This study can be extrapolated to the scientific community at large because characteristics and management of these lesions are similar for our cases although prevalence from other parts of the world differs due to diverse environmental influences [1,2]. The tumors are commoner in males than females as documented in other studies. This is because men are generally more exposed to the risk factors [1-3]. Lipoma has been documented to be the most common mesenchymal tumor and occur in different parts of the body [4], and this was supported by our study. In the orofacial region, cheek has been reported to be the commonest site and this was also corroborated by our study.

We also reported a case of multiple lipoma found in a male patient and it is in conformity with a literature review that stated that multiple lipoma is commoner in males [5]. While solitary is more in females but this is not supported by our study in which we found more solitary lipoma in males than females, this may be due to the fact that with more males growing more fat with alcohol intake and diet changes, especially in abnormal positions, the tendency for benign transformation increases. Although lipoma rarely transform into liposarcoma, both has almost similar histology except for some scattered fibroblasts and signet rings in the malignant lesion [4]. Commonest treatment for lipoma is surgical excision to prevent recurrence but because of scarring,

liposuction is adopted in some extensive or multiple cases and multiple steroid therapy for small lesions [4-8].

Hemangiomas are usually childhood tumor-like growths and it comprises infantile and congenital [9]. However, cherry hemangiomas are bumps that develop on the skin, often later in life, but are not cancerous. Infantile hemangiomas are postnatal, developmental, with biphasic growth characterized by initial proliferations which start after birth and increases at varying rates to different sizes/extents. It is then followed by almost complete involution at the rate of about 10% per year [9]. Normal skin is not always restored in all cases following involution. Congenital hemangiomas are the ones that have attained already large size at birth and two groups exist, the first never involutes but continue to grow into adulthood called; never involuting congenital hemangioma (NICH) and second group rapidly involutes and referred to as rapidly involuting congenital hemangioma (RICH) [10]. Thorough evaluation and continued observation and treatment is individually based [10].

Factors that determine treatment offered for hemangioma include the stage of growth (proliferation vs. involution) of the lesion, complications, and psychosocial requirement. Treatment includes observation, laser therapy, cryotherapy and embolization under CT angiogram guidance. Drug therapy (sclerotherapy) is useful in the proliferative phase, and surgical removal with feeder ligation is applicable especially in the involution phase to minimize bleeding. Combination treatment has also been adopted [10-13]. This study documented 13.3% cases of hemangioma affecting the lips, lids and cheeks and all were treated by feeder vessel ligation and surgical excision with meticulous control of bleeders. Primary closure was achievable in all the cases.

Neurofibroma could present as solitary small or large (elephantiasis) lesion, plexiform, diffuse and it can be multiple as in Von-Recklinghausen's disease or in association with multiple endocrine neoplasia type 3 [14,15]. It is very common in the oral cavity but frequently present in the head and neck region and it originates from the endoneurium of any of the nerves. Schwannoma originate from the nerve sheaths (myelin sheath and Schwann cells). Lesions affecting nerves with bony canals produce the Blunderbuss expansion of the canal [14-16]. The World Health Organization (WHO) has subdivided neurofibroma into 2 broad categories: dermal and plexiform.

Dermal neurofibroma arise from a single peripheral nerve, while plexiform neurofibromas are associated with multiple nerve bundles. Localized neurofibroma (sporadic neurofibroma), diffuse neurofibroma, plexiform neurofibroma, and epithelioid neurofibroma are the other clinicopathological subtypes documented in literature [17-20]. Close to 21% of cases plexiform type which present like a bag of worms have been reported with about 5% having malignant transformation [21]. In our study, we recorded 10% cases out of which 8.3% was solitary types. Surgical excision is done for solitary cases but where lesions are multiple, formation of scars and even recurrence are issue to contend with and the cosmetic and

functional benefits of excision are issues to consider.

Squamous cell carcinoma was the most common malignant epithelial tumor documented in literature and it was commoner in middle aged and elderly, with clinic-pathological types which include verrucous, papillary/exophytic, basaloid, spindle and granular types and, well, moderately, poorly differentiated histological types [22]. Two (3.4%) cases of rhabdomyosarcoma, were documented in our report, one case each of embryonal and pleomorphic types. Three histological types exist and these are embryonal (with multiple variants like butyroides, spindle), alveolar and pleomorphic [23]. The embryonal is more common in the head and neck and in children below 10 years, alveolar type which is the most aggressive, occur in over 10 years while the pleomorphic occur above 30 years.

We documented 13.3% of malignant tumors, majority were stages 1 or 2, except for a case of stage 3 squamous cell carcinoma and a stage 4 malignant schwannoma. The patient with malignant schwannoma had multiple operations. The first operation for the patient was to reconstruct over 70% of the upper and lower cheek as well as half of the upper and lower lips. Four weeks after tumor ablation, closure of the defect was achieved by delayed reconstruction with regional cervical flap for intraoral lining and Bakamjiam (deltopectoral flap for cover and repair of the lips). Patient was not immediately referred for radiotherapy considering the fact that the lesion is not really radiosensitive, but with recurrence/new growths following four excisions, a decision was taken to send for radiotherapy, he was however lost to follow-up. Many other free flaps have been adopted by various surgeons for reconstruction of soft tissue defects and these include latissimus dorsi, radial forearm, collateral arm, lateral thigh free flaps and, when bone was eroded and resected, composite flaps like deep circumflex iliac artery groin flaps or better still fibula flaps with larger vessels and longer bone were used [3]. The tongue can divide into 4 quarters; in addition, lesions can involve the tip, anterior two-third, posterior one-third or base of the tongue either on the lateral, ventral or dorsal surfaces. The lip is subdivided into 3 vertical portions (central and laterals), and 3 horizontal portions which are the attached part of lip, the free part and the vermillion border with the upper and lower lips linked at the commissure.

Malignancies in the floor of the mouth and lingual sulcus usually spread to involve the ventrolateral surface of the tongue [24]. With excisions that involve one-quarter of the tongue and one-third of the lip, and small defects of the cheek, it is possible to close primarily by undermining and there will be minimal aesthetic and functional deficit but with larger defects, other local flap techniques such as Abbe, Eslander, Bernard, Karanpandzic, Gillie's, W-Y flaps or regional pedicled flaps such as supraclavicular fasciocutaneous flaps based on transverse cervical artery are recommended [3,24].

Because of the low socioeconomic status of our patients and lack of viable insurance policy, we had limitations of inability of our patients to afford CT and MRI scans, and nuclear

imaging techniques such as positron emission tomography/ single photon emission computerized tomography (PET/ SPECT) for appropriate staging and follow-up were not yet available. Cervical nodal metastases have been documented as the most important prognostic indicator of head and neck tumors. Sentinel lymph node biopsy using gamma probes attached to analyzer to detect radiations from the sentinel node prior to excision remains a useful means for the diagnosis of occult metastases [25]. Haerle and Stoeckli compared the accuracy of diagnosis of suspicious lymph nodes using planar lymphoscintigraphy and hybrid 3D SPECT/CT scan and concluded that latter was more sensitive and accurate [25]. The treatment for such stage 3 malignancies which we adopted is combination of radical surgery to remove all the tumors at the primary site with adequate safety margin, functional dissection subtypes 1, 2 or 3, and radiotherapy of both primary sites and neck.

In conclusion, we presented a report of soft tissue tumors with a high prevalence of benign tumors than the malignant tumors. All hamartomas/benign tumors were amenable to conventional surgical excision with satisfactory aesthetic and functional outcome. Malignant tumors responded well to the conventional therapy with only one case of multiple recurrences. Facilities for further evaluation and management of these lesions especially the malignant tumors are still inadequate and efforts are already been directed towards acquiring these facilities particularly with the establishment of our nuclear medicine center.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

Authors' contributions	BOA	OGO	OAA
Research concept and design	✓	--	✓
Collection and/or assembly of data	✓	✓	✓
Data analysis and interpretation	✓	--	--
Writing the article	✓	✓	--
Critical revision of the article	✓	✓	--
Final approval of article	✓	✓	✓
Statistical analysis	✓	--	--
Involvement in surgery	✓	--	✓
Involvement in histology	--	✓	--

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