Intraoral pleomorphic adenoma: A report of three cases and review of the literature

Omisakin OO¹, Ajike SO², Ayuba GI³

¹Department of Surgery/Maxillofacial Unit, Kaduna State University, Kaduna, Nigeria. ²Department of Dental Surgery, Ahmadu Bello University, Zaria, Nigeria. ³Department of Anatomic Pathology/Forensic Medicine, Kaduna State University, Kaduna, Nigeria.

*Corresponding author: omisakinolatunde@gmail.com

Received: 22.12.16; Accepted: 16.08.17; Published: 21.08.17

ABSTRACT

Background: Pleomorphic adenoma, a benign salivary gland neoplasm, is characterized by neoplastic proliferation of parenchymatous glandular cells along with myoepithelial components. It is the commonest type of salivary gland tumour and parotid gland tumour. It is usually solitary and presents as a painless, uninodular, firm and mass. Aim: To highlight the clinical features and management of intraoral pleomorphic adenoma. Methods: Three clinical cases of pleomorphic adenoma that occurred in the oral cavity were used to illustrate the presentation and surgical management of the lesion. Findings: The first clinical case was 18-year old girl with a circumscribed mass in the upper lip of 2 years duration. The second case was 18-year old girl with a tumour on the palate of 3 years duration. The third case was 24-year old boy with a bulging tumour of the upper lip of 10 years duration. All the tumours were surgically excised. Conclusion: Diagnosis of pleomorphic adenoma at an early stage makes management effective and prognosis of treatment very good. Recommendations: Recurrence of pleomorphic adenoma had been documented in the literature. Therefore, precise surgical excision is mandatory.

Key words: Pleomorphic adenoma, salivary gland, neoplasm, tumour, oral cavity, teething

INTRODUCTION

Pleomorphic adenoma is the commonest benign salivary gland neoplasms.[1] Over 84% of pleomorphic adenomas occur in the parotid gland, 8% occur in the submandibular gland, 1% occur in the sublingual and 6.4% occur in the minor salivary glands.[1]

Pleomorphic adenomas are usually seen in middle aged women and present as painless
slowly growing mass. These masses are solitary and well demarcated. They may appear heterogeneous, secondary to hemorrhage; calcification and neurosis. This lesion exhibits a wide cytomorphologic and architectural diversity. It is the most common benign tumour of minor salivary glands and the palate is the most common site, then the upper lip followed by the buccal mucosa. Other intraoral sites commonly affected by this tumour are: the buccal mucosa, floor of the mouth, tongue, tonsils, and pharynx and retromolar region.

Intra oral pleomorphic adenoma appears as slowly growing, painless mass usually in the fourth or fifth decade of life. Patients typically present with a smooth painless enlarging mass. Although it is a benign tumor, it has a tendency to be locally aggressive: in few cases it has degenerated into a malignant tumour.

The mainstay of management is wide local excision with the removal of periosteum or bone if they are involved. “Rupture of the capsule or tumour spillage is also believed to increase the risk of recurrence”. The differential diagnosis of intraoral pleomorphic adenoma include: fibroma, lipoma, neurofibroma, palatal torus and neurilemmoma.

Pleomorphic adenoma is a mixed tumour because it has both epithelial and connective tissue elements. “It is composed of epithelial and myoepithelial cells arranged with various morphological patterns, demarcated from surrounding tissues by fibrous capsule”.

The aim of this report is to present the occurrence of pleomorphic adenoma in the oral cavity and its management. This report consists of three cases of pleomorphic adenoma that occurred in the oral cavity, and they occurred during the second and third decade of life.

CASES ONE

An 18-year old girl presented in the dental clinic of Barau Dikko Teaching Hospital, Kaduna with complaint of painless swelling in the upper lip of two years duration. On examination, there was a palpable mobile, hard, multi-lobulated swelling in the substance of the upper lip which was not tender to palpation. The overlying mucosa was normal and not ulcerated but there was a bulging mass showing intra orally (figure1). The mild disfigurement of the face made the patient to seek medical care. The patient was in good state of health. She had no symptoms of any systemic diseases. The patient blood investigations results are shown in table 1.

Figure 1: Tumour bulging from upper lip after incision

Figure 2: The excised tumour
Surgical excision of the tumour was done under local anaesthesia with lidnocaine 1:10,000 dilution of adrenaline. The excised tumour measured 4cm by 3cm. It was nodular and firm in consistency (figure 2). The histology report showed proliferating epithelial cells in form of cuboidal cells in sheets and fibro-collagenous stroma and fibromyxoid tissue within a capsule (figure 3). No invasion of the capsule was noted. Diagnosis of pleomorphic adenoma was made based on these histological features.

**CASE TWO**

An 18-year old female patient reported to the dental clinic of the Barau Dikko Teaching Hospital, with a complaint of painless swelling in the left palatal region of 3 years duration. The tumour affected her speech and swallowing often times. Examination of the oral cavity revealed a round, circumscribed mass at the junction of hard and soft palate on the left side (figure 4). The mass was asymptomatic, slow growing, firm, with smooth surface and no radiographic evidence of bone involvement was seen. The blood investigation result is shown in (table 2). The patient had no symptoms of any systemic diseases. Surgical excision of the tumour was done under general anaesthesia with a margin of 2mm of apparently normal palatal mucosa (figure 5). The excised tumour is well circumscribed measured 2cm X 3cm (figure 6). The histology of the tumour showed myoepithelial cells with trabecular sheets, proliferating epithelial cells in form of cuboidal cells in sheets and fibrocollagenous stroma and fibromyxoid tissue within a capsule (figure 7).

<table>
<thead>
<tr>
<th>PARAMETER</th>
<th>VALUE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Haemoglobin concentration</td>
<td>12gm/dl</td>
</tr>
<tr>
<td>Serum potassium</td>
<td>3.8mmol</td>
</tr>
<tr>
<td>Sodium</td>
<td>145mmol</td>
</tr>
<tr>
<td>Bicarbonate</td>
<td>3.5mmol</td>
</tr>
<tr>
<td>Urea</td>
<td>2.5mmol</td>
</tr>
</tbody>
</table>

**Table 2: Results of blood investigation**

<table>
<thead>
<tr>
<th>PARAMETER</th>
<th>VALUE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Haemoglobin concentration</td>
<td>12.8gm/dl</td>
</tr>
<tr>
<td>Potassium</td>
<td>4.8mmol</td>
</tr>
<tr>
<td>Sodium</td>
<td>142mmol</td>
</tr>
<tr>
<td>Bicarbonate</td>
<td>4.6mmol</td>
</tr>
<tr>
<td>Urea</td>
<td>3.0mmol</td>
</tr>
</tbody>
</table>
CASE THREE

A 24 year old male patient was referred from a peripheral hospital to Maxillofacial Clinic of Barau Dikko Teaching Hospital following a complaint of jaw tumour of ten years duration. The patient claimed to have noticed the swelling about ten years ago in the upper jaw. The tumour had been growing slowly with no associated pain and ulceration. The present appearance of the tumour made the patient to seek medical care. Examination revealed a bulging tumour protruding from the upper lip. The tumour displaced the upper anterior teeth lingually and the teeth were mobile. The tumour was firm, attached to overlying mucosa but not tender to palpation and involve the right alveolar process of the maxillae (figure 8). There was no regional lymphadenopathy. The patient was in a good state of health. Blood investigation results are shown in table 3. Surgical excision of the tumour was done under general anaesthesia.

Prosthetic restoration of the loss alveolar process and teeth was done. The excised tumour measured 7cm X 6cm (figure 9). The histology showed cuboidal epithelial cells, with myxoid component with stroma in between contains dispersed spindled with trabecular, tubular, cystic and papillary architecture (figure 10). A diagnosis of pleomorphic adenoma was made.
Table 3: Blood investigation results

<table>
<thead>
<tr>
<th>PARAMETER</th>
<th>VALUE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Haemoglobin concentration</td>
<td>14.5gm/dl</td>
</tr>
<tr>
<td>Potassium</td>
<td>4.0mmol</td>
</tr>
<tr>
<td>Sodium</td>
<td>150mmol</td>
</tr>
<tr>
<td>Bicarbonate</td>
<td>4.8mmol</td>
</tr>
<tr>
<td>Urea</td>
<td>2.8mmol</td>
</tr>
</tbody>
</table>

DISCUSSION

Pleomorphic adenoma is the commonest benign salivary gland tumour of both major and minor salivary glands, 84% of it occur in the parotid gland, 8% in the submandibular, 1% in the sublingual and 6.4% in the minor salivary glands.[1] It is the most common salivary gland tumour in both children and adults.[2]

Females are more affected than males at ratio of 2:1.[3] The reported cases involved two females and one male. Pleomorphic adenoma occurs in the fourth and fifth decades of life, this may be due slow growing nature of the tumour and usually there is no associated pains, therefore late presentation, but may arise at any age;[4] the three cases reported were in second and third decades of life this may be due to their early presentation to the hospital.

Pleomorphic adenoma is the most prevalent benign tumour of the minor salivary glands.[4] The most common site of this tumour is the palate followed by the upper lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, and retromolar region.[4] Review of literature reported few cases of pleomorphic adenoma arising in the parapharyngeal space.[5] Two of the cases in this report occurred in the upper lip and one on the palate.

Pleomorphic adenoma arises in the oral cavity as a painless, slowly growing, firm swelling, commonly seen on the posterior lateral aspect of the palate, presenting as a smooth, dome shaped mass.[6] It appear fixed on the palate because of the tightly bound nature of the hard palate mucosa.[6] While in the lip and buccal mucosa, it is freely movable.[6] The third case presented here was not mobile but firmly attached to the substance of the lip, this may be due to long duration of the tumour. Pleomorphic adenoma of the palate is rarely allowed to attain a size greater than 1 to 2cm in diameter because it causes difficulty in mastication, speech, and swallowing.[7] It is detected and treated earlier than tumours of major salivary glands.[7] If the overlying mucosa is ulcerated and ulceration is not due to any trauma or biopsy, malignancy should be suspected.[8]

Computerized tomography scan is the most important diagnostic tool of these tumours. It helps to determine the extension of the lesion. Pleomorphic adenoma cannot invade the bone but may lead to resorption of the bone due to pressure effect.[6]

“It is an epithelial tumor of complex morphology; possessing epithelial and myoepithelial elements arranged in varieties of pattern and embedded in mucopolysacharides stroma (mesenchymal component).”[8] The epithelial cells are usually cuboidal with clear or eosinophilic cytoplasm. Stroma in between contains dispersed spindled and epitheloid myoepithelial cells.[6]

The treatment for pleomorphic adenoma is surgical excision.[9] Tumours of hard palate are usually excised down to the periosteum including the overlying mucosa with 1cm clinical margins at the periphery.[10]

Pleomorphic adenoma does not usually recur after adequate surgical excision.[9] “Causes of recurrence include incomplete excision, seeding, cutting through microscopic extracapsular projections thereby leaving some tumour behind, or rupture of the capsule and accidental seeding of tumor cells”.[8,9]

The three cases reported were followed up for 10 months. The healing has been satisfactory with no complaints and no sign of recurrence.

“Three histological types have been described: Myxoid (hypocellular) which is most common, then cellular and classic.”[6] There is a minimal risk of malignant transformation into a carcinoma ex-pleomorphic adenoma, which is usually associated with the time the lesion is in situ (1.5% in first 5years, 9.5% after 15years). Excision is recommended essentially in all cases.[10]
“Additional risk factor for malignancy includes advanced age, large size, radiation therapy and recurrent tumours. In addition to carcinoma ex-pleomorphic adenoma, true malignant mixed tumour of the salivary glands usually arise from pre-existing pleomorphic adenoma”. [10,11,12] “Metastasizing pleomorphic adenoma is the third type of malignant mixed tumour of salivary glands and is the rarest” [10,13,14]. “It present with metastases to lungs, bone and soft tissues despite having a benign histology.” [10]

CONCLUSION

Pleomorphic adenoma of minor salivary glands have very good prognosis after excision. Surgical excision should not be delayed after diagnosis.

RECOMMENDATION

Tumours involving the oral cavity needed to be treated urgently to avoid pressure resorption of associated bony structures.

REFERENCES


How to cite this article: Omisakin OO, Aijke SO, Ayuba GI. Intraoral pleomorphic adenoma: A report of three cases and review of the literature. Int J Med Biomed Res 2017;6(2):59-64

Conflict of Interest: None declared
Submit your next manuscript to any of our journals that is the best fit for your research

International Journal of Medicine and Biomedical Research
Scope: UMBR publishes cutting edge studies in medical sciences
Editor-in-Chief: Sofola A. Olusoga, MBBS, PhD, FAS
Deputy Editor: Lehr J. Eric, MD, PhD, FRCSC
URL: www.ijmbr.com
E-mail: editor@ijmbr.com
Pissn: 2277-0941, eISSN: 2315-5019

International Journal of Ethnomedicine and Pharmacognosy
Scope: IEP publishes novel findings on the use of complementary and alternative medicine in the management of diseases
Editor-in-Chief: Dickson A. Rita, B.Pharm, GCAP, PhD, MPSGh, MCPA
Deputy Editor: Kuete V., PhD
URL: www.ijepharm.com
E-mail: editor@ijepharm.com
Pissn: 2437-1262, eISSN: 2437-1254

International Journal of Infectious and Tropical Diseases
Scope: IITD publishes interesting findings on infectious and tropical diseases of public health importance
Editor-in-Chief: Yang Z., PhD
Deputy Editor: Liping L.P., MD, PhD
URL: www.ijitd.com
E-mail: editor@ijitd.com
Pissn: 2384-6607, eISSN: 2384-6585

Reasons to publish your manuscript with Michael Joanna Publications:
- User-friendly online submission
- Rigorous, constructive and unbiased peer-review
- No space constraints or coloured figure charges
- Immediate publication on acceptance
- Authors retain copyright
- Inclusion in AJOL, CAS, CNKI, DOAJ, EBSCO, Google Scholar, and J-Gate
- Unlimited and wide readership
- Member of COPE and CrossRef

Editorial Director
Professor Sofola A. Olusoga,
Department of Physiology,
University of Lagos,
Nigeria.
Tel: +234(0) 7093848134
Email: enquiry@michaeljoanna.com
www.michaeljoanna.com