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RESEARCH ARTICLE

PRIMARY TUMORS OF THE SUBMANDIBULAR GLAND: A REPORT OF 20 CASES

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Abstract

Salivary gland tumors are rare, accounting for 3% of all tumors in the body and 6% of tumors in the head and neck. Tumors of the submandib ular gland are less common than those of the parotid gland and present both diagnostic and therapeutic challenges, particularly regarding the indication for postoperative radiotherapy and the management of recurrence. The aim of our study was to analyze the epidemiological, clinical, paraclinical, therapeutic, and prognostic features of submandib ular gland tumors based on a retrospective study including a series of 20 cases of submandibular gland swelling managed in the departments of ENT and Head and Neak Surgery at the Nouakchott National Hospital (CHN) from January 2019 to October 2023. The study involved 8 men and 12 women. The average age was 37.9 years (range: 20-71 years). Submandibular swelling was the main symptom. Pain was reported by 14 patients. One case of paralysis of the mental branch of the facial nerve was observed during clinical examination. In terms of treatment, submandibulectomy was performed in 19 patients. Histopathological results showed a predominance of pleomorphic adenoma among benign tumors (70% of benign tumors) and two cases of submandibular tuberculosis (10%). Two benign tumors that recurred were both pleomorphic adenomas.Submandibular gland tumors are complex, and predominantly benign. They represent diagnostic and therapeutic issues. Diagnosis is guided by clinical and radiological evidence and is based on the histopathological analysis of the surgical specimen.

Introduction:-

Tumors of the submandibular region are a relatively rare pathological entity, characterized by a uniform set of clinical features[1]. Salivary gland tumors are uncommon accounting for 3% of head and neck tumors and 0.6% of all human body tumors. Tumors of the submandibular gland are less common than those of the parotid gland, accounting for 5-15% of all salivary gland tumors. They pose as many diagnostic and therapeutic challenges, especially regarding the indication of postoperative radiotherapy and the management of tumor recurrence[2,3,4,5,6,7,8].

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These tumors are characterized by a wide histological diversity, but malignant forms predominate[4, 9, 10, 11] with adenoid cystic carcinoma being the most common, followed by mucoepidermoid carcinoma and squamous cell carcinoma. Pleomorphic adenoma dominates the benign histologic subtypes [12, 13, 14].

The most frequent presentation is the appearance of a swelling in the submandibular space.
Additional tests, particularly imaging and fine-needle aspiration cytology for some teams may provide some evidence in favour of the aetiological diagnosis. definitive diagnosis requires submandibulectomy followed by histopathological examination.

Treatment for these tumors is currently mainly surgical and may sometimes be combined with radiotherapy, as chemotherapy has not yet proven effective[15].
However, these tumors pose many diagnostic and therapeutic problems, particularly regarding the appropriateness of postoperative radiotherapy and the management of recurrences[12, 14].
The major surgical complication remains nerve damage, mainly paralysis of the mental branch of the facial nerve[12, 14,16].

The aim of this retrospective study is to compare our epidemiological, clinical, paraclinical, therapeutic, and outcome results for this pathology in a series of 20 patients within the ENT and Head and Neck Surgery Department of the Nouakchott National Hospital between 2019 and 2023 with data from the literature.

Methods:-

This was a retrospective study of patients with submandibular tumors treated in the ENT and Head and Neck Surgery Department between 2019 and 2023.

Results:-

Epidemiology
Sex Our patients consisted of 8 men and 12 women. The male-to-female ratio was 0.6. Age

In our series, the mean age of the patients was 39.7 ± 20.7 years, with a range from 13 to 84 years. The distribution of patients by age group is shown in (Figure1). We noted 3 cases of patients under 20 years of age.

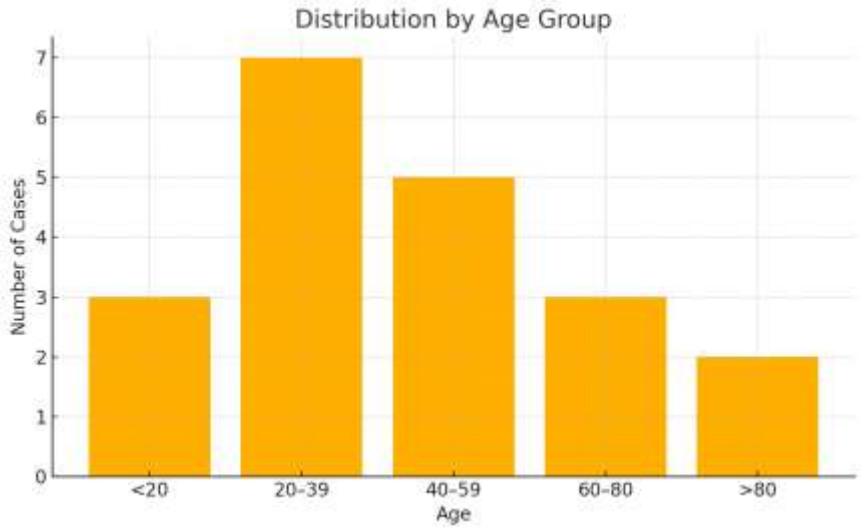

Figure 1: Distribution of patients by age group

Geographic Origin
Patients were most frequently from Nouakchott

Medical History
Patient Interview :
History of smoking in 6 patients
History of pulmonary tuberculosis in 2 patients
History of renal lithiasis in 3 patients
One patient was on long-term medication

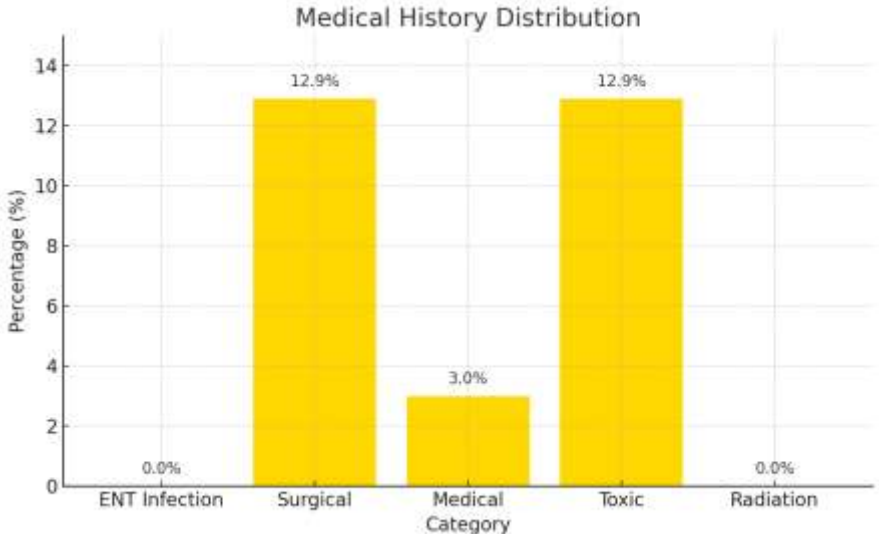

Figure 2: Distribution of patients by medical history

Tableau I : Histological classification of patients

Histological Type	Number (n)	Frequency(%)
Pleomorphicadenoma	14	**70%**
Tuberculosis	2	**10%**
Lithiasis	3	**15%**
Non-conclusive	1	**5%**
Total	**20**	**100%**

Consultationdelay
The mean consultation delay, defined as the time between symptom onset and first medical evaluation was 5.6 ± 3 months (ranging from 15 days to 2 years).

Clinical presentation
Submandibularswelling in all 20 patients
5 patients reportedsalivarycolic
2 patients reportedXerostomia
1 patient had reflex otalgia
The overlying skin appeared normal in 15 patients and abnormal in 5 patients
No patient presented with mental branch palsy of the facial nerve

Physical examination
Palpation of the submandibular swelling
Submandibular swelling is the common presenting symptom in all our patients. The mass was painless, with regular contours, and mobile in both planes.

Intra-oral examination

The systematic examination of the oral cavity yielded the following results:

- The vestibular mucosa was congested in 10 patients
- Vestibular swelling was noted in all patients
- A pronounced budding appearance was observed in 2 patients, and the remaining patients presented withnodular lesions
- No facial sensory disturbances were observed in our patients
- The oral health and dental hygiene was poor in 40% of cases, fair in 40% of cases, and good in the remaining 20%

Neurologicalsigns

All of our patients retained facial sensation and motor function of the mimic muscles

Examination of lymphnode areas

Only one of our patients had a 1 cmx1 cm mobile and painless cervical lymph node. Upon admission, none of the patients had palpable cervical lymph nodes

Radiological findings

Cervicalultrasoud

Ultrasound was performed on all our patients:

- Hyperechoic calcified images were noted in three patients (salivary lithiasis)
- A roughly rounded, well-defined, tissue-echoic image was observed in 10 patients (pleomorphic adenoma)
- Two patients presented with a heterogeneous appearance with slight glandular hypertrophy
- One patient showed homogeneous hypertrophy.

Figure 3: Cervical ultrasoud showing a right submandibular gland sialolithiasis

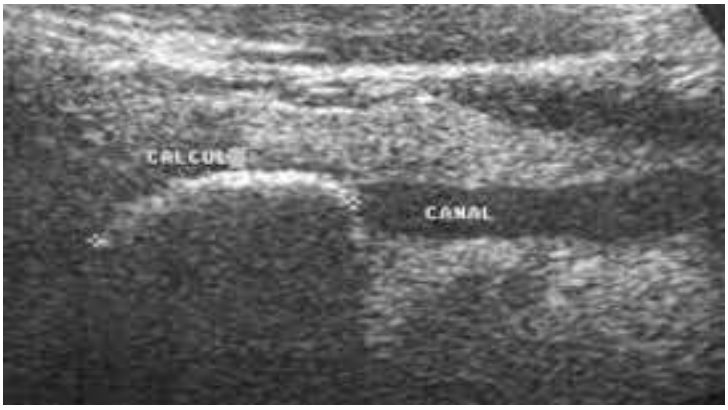

Figure 4:Cervical ultrasound revealing a fluid collection in the left submandibular gland suspicious

Maxillofacial CT scan
Performed in 2 patients, revealing submandibular lithiasis

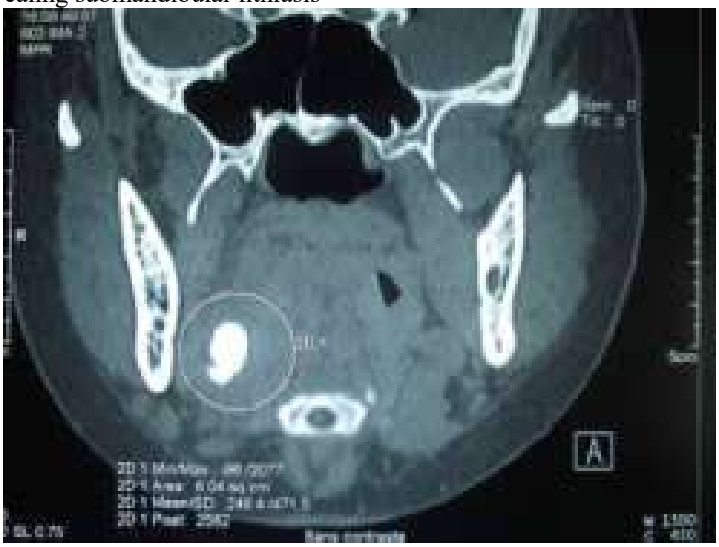

Figure 5:Axial CT of the cervicofacial region showing right mandibular gland sialolithiasis

Maxillofacial MRI
Not performed in our patients

Therapeutic management
Surgery
Therapeutically, submandibulectomy was performed in 19 patients (95%), including 14 operated on for pleomorphic adenoma, 3 for lithiasis, and 2 for tuberculosis of the submandibular gland.

No lymph node dissection was performed in any of our patients.

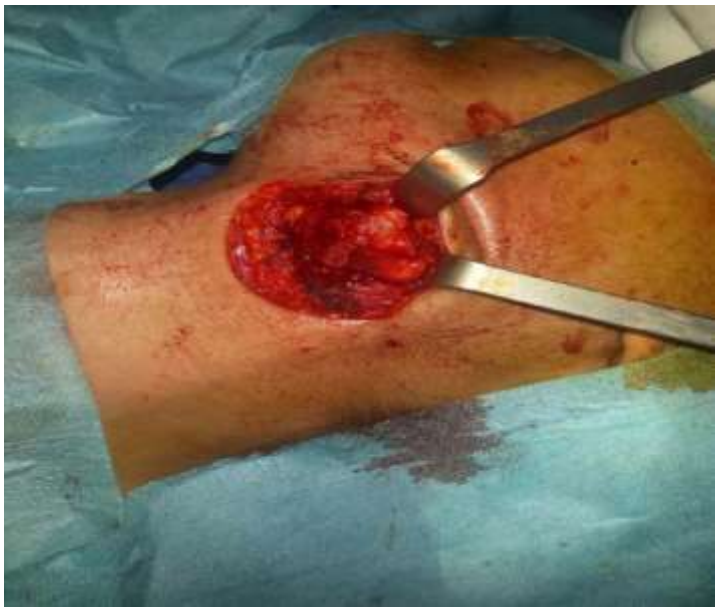

Figure 6 :Surgical approach for submandibulectomy

Medical treatment
One patient (5%) was treated with corticosteroids.
Patients whose histology confirmed tuberculosis were referred to a specialized center to begin anti-tubercular treatment. Clinical follow-up showed a marked improvement in symptoms, restoration of general condition and weight gain.

Radiotherapy and Chemotherapy
No patient in our series required radiotherapy or chemotherapy.

Histopathological findings
Histological examination revealed a predominance of pleomorphic adenomas in 14 patients (70%) and two patients with tuberculosis (10%).

Therapeutic results
All our patients were followed up postoperatively, and their progress was assessed mainly based on clinical criteria, through a complete clinical examination, particularly of the submandibular space, face, oral cavity, and cervical lymph node areas. Follow-up was scheduled as follows, depending on the patient:
- One month postoperatively
- Threemonths
- Six months
- And then at 12 months

Evolution
II.11.1 ImmediatePostoperative Complications
II.11.2 Facial Nerve Function:
Facial function analysis was evaluated after excluding two patients who had pre-existing facial paresis.

- Facial involvement in these cases was predictable postoperatively. Facial dysfunction in the immediate postoperative period was paresis in two patients: One case of them developed mental branch paresis following a surgical reintervention
- Patient age was not correlated with facial nerve impairment after submandibulectomy (Chi2 test not significant, p=0.23), nor was the type of lesion and nerve injury (Chi2 test not significant, p=0.11).

- One Hypoglossal nerve injury was observed

II.11.3 Hemorrhage and/or Hematoma:
No cases of postoperative hemorrhage or hematoma were observed in our patients.

II.12 Short and Mid-Term Evolution
- At one month (M1), facial paresis persisted
- At three months (M3), full recovery was noted in the marginal mandibular branch territory
- At six months (M6), there were no remaining cases of facial paresis
- Among the postoperative complications, we noted two cases of submandibular depression, one case of salivary fistula which resolved well after one month, and no cases of surgical site infection.

II.13 Long-Term Evolution and Sequelae:
The outcome in our patients was good after 12 months of follow-up.
No cases of Frey's syndrome were observed; however, two cases of submandibular depression persisted, considered as sequelae.
No deaths were reported in our series, and there were no cases of recurrence.

II.14 Management of post-operative complications:
Patients who experienced postoperative facial paresis received a corticosteroid bolus followed by oral medication, artificial tears, and facial physiotherapy. The salivary fistula resolved with local care combined with oral antibiotics.

Discussion:-

Salivary gland tumors are rare, accounting for 3% of head and neck tumors and 0.6% of all human neoplasms[4, 5]. Approximately 10% of all salivary gland tumors are located in the submandibular gland, with a high rate of malignancy. The average age of discovery of submandibular tumors is between the 4th and 5th decades of life. The overall sex ratio varies according to studies, with a slight predominance in females, consistent with our study [1, 2, 5, 6]. ubmandibular gland tumors usually present as a painless, slow-growing swelling. Malignancy should be suspected in the presence of a firm, painful submandibular swelling that is more or less fixed at the superficial and/or deep level. The presence of palpable satellite lymphadenopathy and skin, bone, or nerve involvement should raise suspicion of possible locoregional extension. However, none of these signs are absolute, and the distinction between benign and malignant remains challenging[6, 7]. Imaging plays a huge role in providing anatomical and morphological information, although it lacks specificity. Some authors defend the value of ultrasound, especially in differentiating between benign and malignant tumors [3]. Malignant features are mainly heterogeneous, poorly defined masses, occasionally with extra glandular extension and sometimes associated with lymphadenopathy [7]. In our study, ultrasound did not suggest a malignant tumor, revealing a hypoechoic mass with irregular contours, nor did the CT scan, which could be associated with ipsilateral cervical lymphadenopathy. Other authors consider that CT remains the method of choice, particularly for determining the locoregional extent of the disease, given its availability and the wealth of information it provides [8]. MRI, with its ability to explore soft tissue in three dimensions, offers excellent topographic delineation of tumors relative to vascular and neural structures and is especially useful for assessing recurrence [3, 8]. However, MRI was not performed in our patients due to financial constraints.

In this study, only histopathological analysis of the surgical specimen can confirm the definitive diagnosis. Other benign tumors such as salivary lithiasis and submandibular gland tuberculosis account for 15% and 10% of cases, respectively. Most authors agree that malignancy is more common in submandibular lesions than in parotid tumors; with reported rates ranging from 40% to 55% [1, 3, 4]. No cases of malignancy were found in our series.

The treatment of submandibular tumors has been controversial for many years with regard to the type of lesion excision, the approach to lymph node areas, and the role of adjuvant radiotherapy for malignant tumors. The goal of any surgery is to ensure wide excision to reduce the recurrence rate while preserving nerve structures [2, 3]. The main principle of surgery for benign tumors, particularly pleomorphic adenoma, is to perform an external submandibulectomy, which reduces the risk of recurrence and malignant transformation. In the case of a primary malignant tumor, a submandibulectomy and lymph node dissection must be performed [9], due to the high incidence of histologically proven nodal involvement (53%) [1, 4, 9]. he complications of submandibulectomy are dominated by damage to the mental branch of the facial nerve (10%) and to the hypoglossal nerve (5%). Two patients in our series were diagnosed with tuberculosis and received anti-tuberculosis treatment.Long-term monitoring based on clinical examination and imaging is necessary to detect recurrence[4, 10].

Conclusion:-

Submandibular tumors are relatively rare and include a wide variety of histological entities. Radiological assessment is essential, as it often provides clues toward diagnosis, which must then be confirmed by histopathological examination, either an extemporaneous histological examination or through analysis of the surgical specimen.
The therapeutic approach depends on whether the tumor is benign or malignant.
The surgical technique must be adapted to each histological type. Current treatments and the development of minimally invasive techniques assisted by radiography, such as lithotripsy or sialendoscopy, are increasingly used in developed countries with few side effects.

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