Management of Pleomorphic Adenoma of Submandibular Gland: A Rare Case

Elvita Nora Susana[1,2*], Lily Setiani[1,2], and Baluqia Iskandar[1,2]

[1]*Otorhinolaryngology Head and Neck Surgery Department Faculty of Medicine, University of Syiah Kuala*
[2]*dr. Zainoel Abidin Hospital, Banda Aceh, Indonesia*
**Corresponding author: Email: elvitasusan@gmail.com*

ABSTRACT
Pleomorphic adenoma is the most common benign tumor in the salivary glands but rarely in the submandibular glands. It is a mass in the submandibular, which is asymptomatic and painless. Excision of the tumor is a treatment that surgeons widely use. The study aimed to describe the management of pleomorphic adenoma of the submandibular gland. A 55-year-old woman with a lump on the left submandibular since 20 years ago. The result of the Fine Needle Aspiration Biopsy (FNAB) revealed a follicular neoplasm and on a neck computed tomography (CT) scan showed a heterogeneous mass on the left cervical with a size of 7x5,3x7 cm. Incision and transcervical excision of the left submandibular were performed. It started with a horizontal incision that ±11 cm long following the size of the tumor and according to the skin line. This incision was made ±3 cm below the left mandibular angle to avoid the marginal mandibular of the facial nerve branch. The histopathological result was a pleomorphic adenoma. The literature and evidence were searched using Medline, PubMed, and hand searching. Excision of the tumor is the appropriate treatment for pleomorphic adenoma and there are several surgical approaches to the submandibular gland, including transcervical, submental, retroauricular, and transoral lateral. The pleomorphic adenoma submandibular gland is a rare case. Excising the whole pleomorphic adenoma of the submandibular gland and its capsule showed a good result.

KEYWORDS: PLEOMORPHIC ADENOMA, SUBMANDIBULAR GLAND, SURGERY

1. INTRODUCTION

Salivary gland tumors represent 2-6% of all head and neck tumors [1]. Da Silva et al. reported that pleomorphic adenoma was the most common benign tumor (82.2%) in the parotid (50%), palate (25%), submandibular (13.6%), and sublingual (0.36%) glands, with a male to female ratio of 1:1.5 and mean age of 46 years old [2]. The submandibular gland is the second largest salivary gland after the parotid gland. The etiology of pleomorphic adenoma in the salivary glands is unknown, presumably due to environmental and genetic involvement. Radiation is considered a risk factor for the development of salivary gland neoplasms [3,4].

Small pleomorphic adenomas are asymptomatic masses that are solid, mobile, smooth surface, and well-demarcated tumors. Meanwhile, the larger tumor is multinodular and has a rough surface that can make the skin or mucosa overlying thinner and weaker. It can be gray with sizes ranging from 2-5 cm to massive, almost 50 cm in diameter, and 6 kg in weight have been reported [5,6].

Fine Needle Aspiration Biopsy (FNAB) of salivary gland tumors is a simple and low-risk examination that supports the diagnosis of salivary gland neoplasms. Mete et al reported that FNAB had sensitivity (80.8%), specificity (95.1%), false positive (4.9%) and false-negative (19.2%) [7]. Ultrasound can differentiate between normal glandular parenchyma, inflammation, tumors, lymph nodes, and calculi. FNAB with ultrasound guidance provides more accurate results.[6] CT scans and MRI give a better visualization of the salivary gland mass. For minor salivary lesions, CT scan provides information in planning treatment, showing the connection of the tumor to surrounding anatomical structures and tumor extension, also assessing the possibility of severe lesions resection. CT scan is very useful in assessing bone erosion, while MRI demonstrates tumor extension along the cranial nerves [8].

One of the treatments of submandibular gland benign or malignant neoplasms is surgery. In pleomorphic adenomas, excision of the tumor is the appropriate treatment. Entire nerves are preserved unless involved with a tumor [3].

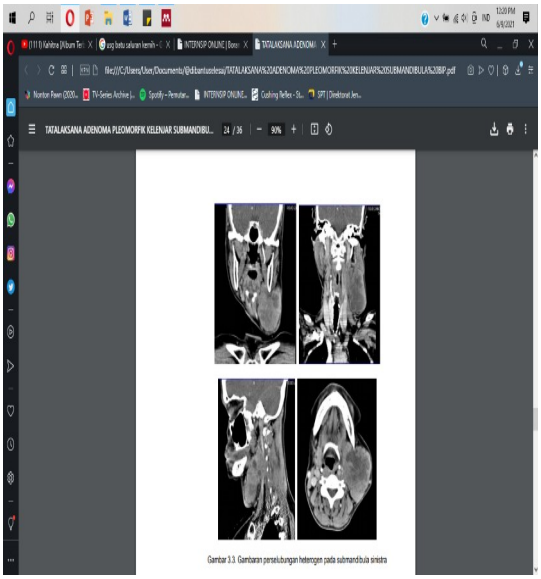
2. METHOD

The literature search was conducted on October 3rd 2019, with the keywords "pleomorphic adenoma" AND "Submandibular gland excision". Results of research on Sciencedirect and PubMed to search for the evidence.

The literature search was performed using the following inclusion criteria: 1) Submandibular pleomorphic adenoma; 2) Excision approaches for submandibular tumors and exclusion criteria as follows: Pleomorphic adenoma originated from other than submandibular gland.

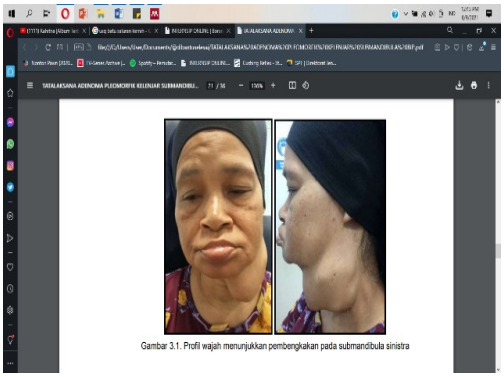
3. RESULT

A Female, Mrs. R, 55 years old, came to the ENT Polyclinic of RSUD dr. Zainoel Abidin Banda Aceh has had a chief complaint of a lump under her left submandibular since 20 years ago. It was small in size of a green bean but gradually grew to an adult fist, especially in the last 3 months, so she decided to get treatment. There was no pain in the lump. There were no hoarseness, shortness of breath, difficulty swallowing, nasal congestion, headache, double vision and history of bleeding. History of palpitations, fatigue, and weight loss was denied. A history of radiation exposure was denied. History of diabetes mellitus and hypertension was denied. The patient came along with the result of the FNAB and Colli CT Scan that had been done previously.

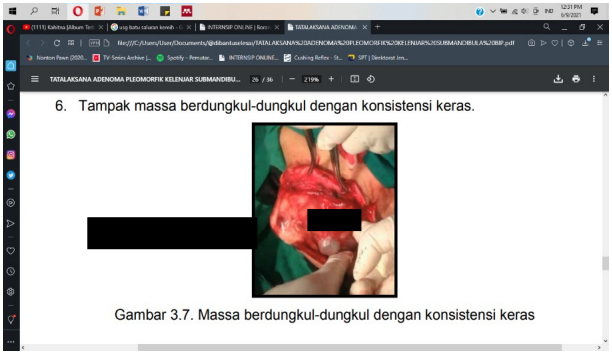


Gambar 3.3. Gambaran penselubungan heterogen pada submandibula sinistra

General status was obtained moderate state, compos



Gambar 3.1. Profil wajah menunjukkan pembengkakan pada submandibula sinistra



Gambar 3.7. Massa berdungkul-dungkul dengan konsistensi keras

anesthesia with ASA III. On May 21st 2019, a left submandibular tumor excision was performed.

Figure 3 (a) Left submandibular incision; (b) Platisma muscle; and (c) A pedunculated rough mass with a hard consistency.

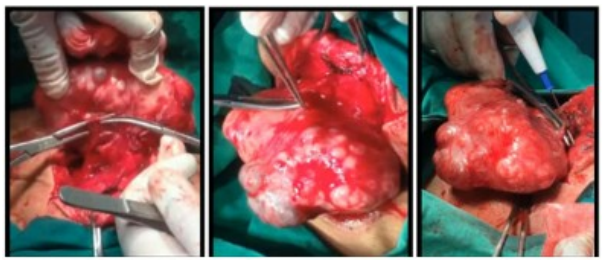


Figure 4 Exempting the edge of the tumor, it appeared that the base of the tumor originates from the submandibular.

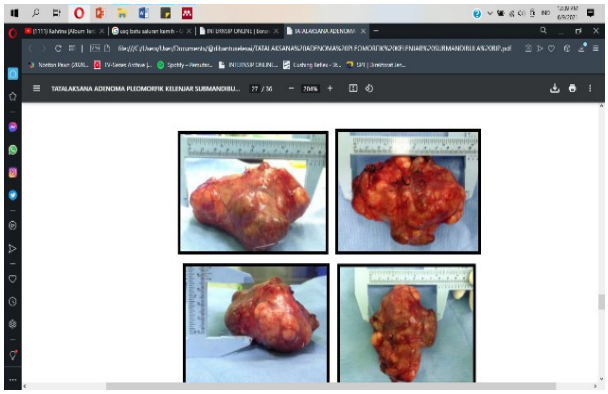


Figure 5 Tumor Size 10 x 8 x 4 cm.

After the surgery, the tumor tissue that had been removed was sent to the Anatomical Pathology laboratory. On the first day follow-up after surgery, the patient complained pain in the surgical wound but vital signs were within normal limits, no blood on the bandage, and blood volume in the drainage was 20 ml.

On the 3rd day after surgery, the pain in the surgical wound was reduced, vital signs were within normal limits, also there was no blood seepage on the bandages covering the surgical wound. On the 10th postoperative day, the patient came to ENT department and there was no any complain, the wound was completely healed, there was no inflammation. Postoperative histopathology examination results showed pleomorphic adenoma.

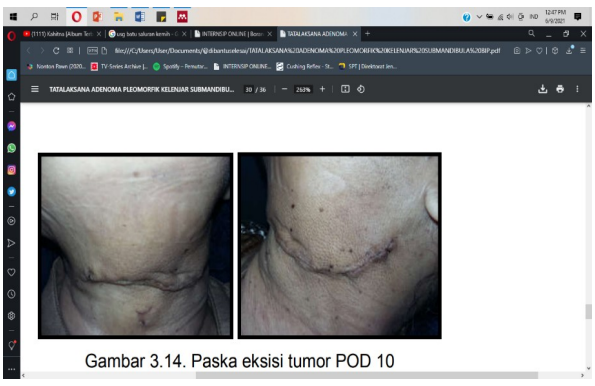


Figure 6. Ten Days after Surgery.

Tumors involving the submandibular gland are often restricted to the gland. In pleomorphic adenomas, excision of the tumor is the appropriate treatment [3]. Beahm et al. described several surgical approaches to the submandibular gland, including transcervical, submental, retroauricular, and transoral lateral [9].

The surgical procedure was performed under general anesthesia. The patient was in the supine position with the head hyperextended. A skin incision is made following the skin fold line about 4 cm below the lower edge of the horizontal portion of the mandibular. It will avoid injury to the mandibular branch of the facial nerve as well as produce a good cosmetic result. An incision of the platysma muscle was made to the fascia. Flaps were made up and down and then made taegel. The release of the tumor starts from the inferior part and then the hypoglossal nerve can be identified. When the anterior margin is freed, the mylohyoid muscle is gently pulled with the retractor and the lingual nerve is visible. After the submandibular gland is removed, the hyoglossus muscle appears as the floor of the submandibular triangle. Penrose drain was installed, then the platysma muscle and skin were stitched up again [10].

4. DISCUSSION

Reported a case of a 55 year old female diagnosed with Submandibular pleomorphic adenoma. Diagnosis is made based on history, physical examination, also radiological and histopathological investigations.

The etiology of pleomorphic adenoma in this patient is unknown. Based on the literature, radiation is considered as a risk factor for the development of salivary gland neoplasms. One study showed that Simian virus (SV 40) played an important role in the development of pleomorphic adenomas. Whereas, Epstein-Barr virus was also a factor in the development of salivary gland lymphoepithelial tumors [3,4].

In this case, the patient complained of a lump 20 years ago in the left submandibular area, which has grown to an adult fist. There is no pain on the lump, it is looked as pedunculated surface, hard consistence, well demarcated, mobile, and with a size of 10x8x4 cm. Literature stated that pleomorphic adenomas appeared as asymptomatic masses with slow growth and in large tumors have an uneven or rough surface and multinodular [5].

Investigations performed on the patient were FNAB and Colli CT Scan with contrast. The FNAB showed a follicular neoplasm, while the histopathology of the postoperative tumor was a pleomorphic adenoma. The literature explains that FNAB has a sensitivity rate of

29-97%. Mete et al. reported that FNAB had sensitivity (80.8%), specificity (95.1%), false positive (4.9%), and false negative (19.2%) [8].

In pleomorphic adenomas, excision of the tumor is the appropriate treatment. In this case, a transcervical tumor was excised with an incision made ±3 cm below the left mandibular angle to avoid the marginal mandibule of the facial nerve branch. The horizontal incision is ±11 cm long following the size of the tumor and according to the skin line. Then the platysma muscle is incised. It looked like an encapsulated tumor with a hard and reddish consistency. The tumor edge was liberated, and the tumor's base was seen from the left submandibular.

The patient was hospitalized for five days and on POD 3 the patient went home for outpatient treatment. Complications that can occur after excision of submandibular gland tumors are trauma to the marginal mandibular branch of the facial nerve which can cause temporary or permanent weakness on the corners of the mouth. Bleeding from the facial artery can also occur after surgery, so exploration and control of bleeding is necessary. Infection after excision of the submandibular gland is rare [3,11]. During hospitalization, evaluation of bleeding through the drain tube and postoperative complications were evaluated. There were no complaints experienced by the patient. Weakness of the lips and numbness of the tongue is absent.

The recurrence rate ranges from 0.8% - 6.8%. Complete excision of the tumor along with the capsule is recommended to reduce the recurrence rate.[6] In this case, excision of the tumor was carried out by removing the whole tumor and its capsule, so hopefully, there would be no recurrence in this patient.

5. CONCLUSION

Pleomorphic adenoma submandibular gland is a rare case. Excising the whole pleomorphic adenoma of submandibular gland and its capsule showed a good result.

CONSENT

The picture and case in this manuscript have been gotten permission from the patient.

REFERENCES

[1] Alshagroud R, Kamoh A, Popat SR, Brandwein-Weber M, Aguirre A. Metastasizing Pleomorphic Adenoma Case Report and Review of the Literature. Head Neck Pathol [Internet]. 2017 Dec 25;11(4):487-93. Available from: http://link.springer.com/10.1007/s12105-017-0815-0
https://doi.org/10.1007/s12105-017-0815-0

[2] Da Silva LP, Serpa MS, Viveiros SK, Sena DAC, de Carvalho Pinho RF, de Abreu Guimarães LD, et al. Salivary gland tumors in a Brazilian population: A 20-year retrospective and multicentric study of 2292 cases. J Cranio-Maxillofacial Surg [Internet]. 2018 Dec;46(12):2227-33. Available from: https://linkinghub.elsevier.com/retrieve/pii/S1010518218304682
https://doi.org/10.1016/j.jcms.2018.09.028

[3] Oh Y, Russell M, Eisele W. Salivary gland neoplasms. In: JT J, CA R, editors. Bailey's Head & Neck Surgery Otolaryngology. 2nd ed. Philadelphia: Lippincott William & Wilkins; 2014. p. 1760-87.

[4] Farhat F, Asnir RA, Yudhistira A, Daulay ER, Sagala IP. An Uncommon Occurrence of Pleomorphic Adenoma in the Submandibular Salivary Gland: A Case Report. Open Access Maced J Med Sci [Internet]. 2018 Jun 17;6(6):1101-3. Available from: https://spiroski.migration.publicknowledgeproject.org/index.php/mjms/article/view/oamjms.2018.248
https://doi.org/10.3889/oamjms.2018.248

[5] Peel R, Seethala R. Pathology of salivary gland disease. In: Myers E, Ferris R, editors. Salivary Gland Disorders. New York: Springer; 2007. p. 45-7.

[6] Homer J, Robson A. Benign salivary gland tumours. In: Watkinson J, Clarke R, editors. Scott-Brown's Otorhinolaryngology Head & Neck Surgery. 8th ed. Boca Raton: CRC press; 2019. p. 115-30.

[7] İnançlı HM, Kanmaz MA, Ural A, Dilek GB. Fine Needle Aspiration Biopsy: in the Diagnosis of Salivary Gland Neoplasms Compared with Histopathology. Indian J Otolaryngol Head Neck Surg [Internet]. 2013 Jul 15;65(S1):121-5. Available from: http://link.springer.com/10.1007/s12070-012-0608-4
https://doi.org/10.1007/s12070-012-0608-4

[8] Shah J, Patel S, Singh B, Wong R. Jatin Shah's Head and Neck Surgery and Oncology. 5th ed. Philadelphia: Elsevier; 2020. 557-608 p.

[9] Beahm DD, Peleaz L, Nuss DW, Schaitkin B, Sedlmayr JC, Rivera-Serrano CM, et al. Surgical approaches to the submandibular gland: A review of literature. Int J Surg

[Internet]. 2009;7(6):503-9. Available from: https://linkinghub.elsevier.com/retrieve/pii/S1743919109001289
https://doi.org/10.1016/j.ijsu.2009.09.006

[10] Medina J, Lore J. The Neck. In: Lore J, Medina J, editors. An Atlas of Head & Neck Surgery. 4th ed. Philadelphia: Elsevier Saunders; 2005. p. 828-31.

[11] Jose M, Desai K. Fatal Superimposed Bacterial Sepsis in a Healthy Coronavirus (COVID-19) Patient. Cureus. 2020;12(5). https://doi.org/10.7759/cureus.8350