

A Rare Case of Ranula on Ventral Surface of the Tongue

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Abstract

Neonatal ranula is one of the head and neck benign abnormal tissue and admitted as a rare case. Neonatal ranula is usually asymptomatic until its size reach threshold to cause functional disturbance or even fatal risk. The lesion which appeared in infant makes a challenge in planning the treatment and needs multidisciplinary involvement to achieve the best result. A 2-months old male infant presented with chief complaint of progressive swelling on ventral side of the tongue since birth. The patient had feeding disturbance and the parents wanted the mass to be removed. The patient had slightly systemic issues based on result of laboratory examination so the surgery was conducted after preoperative tolerance from pediatrician.

The histopathological examination reveals neonatal ranula as histopathological diagnosis. Two weeks after surgery, patient can breastfeed normally. This case illustrates the value of accurate diagnosis and planned management using multidisciplinary approach.

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Introduction

Ranula comes from the Latin word *rana* meaning "frog" because the lesion resembles a bulging stomach of the frog.¹ Theoretically, disruption of the flow of the saliva from the salivary glands leads to the formation of ranula.² The alteration in salivary glands can be divided into two types, extravasation type which caused by the extravasation of mucus after trauma to salivary gland and retention type which caused by partial or complete obstruction of the ducts.³ Most ranula cases occurred in the area of sublingual salivary gland on the floor of the mouth.^{1,3}

Neonatal ranula is a rare case which may resemble choristomas, epidermoid cysts, and lymphangioma.⁴⁻⁶ Ranula are usually

asymptomatic, mostly patients seek treatment after there is significant functional disturbances, such as feeding difficulties and airway obstruction in infants.⁴ Wherefore obstruction of the airway can lead to the fatal risk, treatment need to be established immediately. However reviewing the infants systemic condition to undergo surgical therapy is still necessary. This report presents a rare case of neonatal ranula on the ventral side of the tongue.

Case Report

A 2-months old male infant referred to the Department of Oral and Maxillofacial Surgery of Persahabatan National Hospital, Jakarta, Indonesia from Pediatric Department Cipto Mangunkusumo National Hospital, Jakarta, Indonesia with chief complaint of swelling at his tongue growing slowly since he was born which caused disturbance in breastfeeding. Extra oral examination is within normal limits. Intra orally, there were swelling at the ventral of the tongue with color similar with surrounding mucosa and cystic in consistency (Figure 1), also positive illumination. Patient had no history of fever, vomit, and diarrhea.

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Laboratory examination revealed condition of neutropenia, lymphocytosis, erythrocytopenia, slightly decreased hematocrit counts, slightly elevated prothrombin time, elevated SGOPT count, and decreased ureum count (Table 1). Thorax radiographic examination showed there is tendency the right lower lung and bilateral middle lung contained infiltrates.

discuss several treatment alternatives with the parents and decided to perform extirpation of the mass with general anesthesia.

Test	Result	Unit	Range
Hematology			
Leukocyte	10,17	10 ³ /mm ³	6 ~ 17,5
Basophils	0,2	%	0 ~ 1
Eosinophils	2,8	%	1 ~ 5
Neutrophils	9,0	%	17 ~ 60
Lymphocytes	82,1	%	20 ~ 70
Monocytes	5,9	%	1 ~ 11
Erythrocytes	3,49	10 ⁹ /mL	4,08 ~ 6,05
Hemoglobin	9,8	g/dL	9,5 ~ 13,5
Hematocrit	28	%	29 ~ 41
MCV	79,7	fL	74 ~ 108
MCH	28,1	pg	25 ~ 35
MCHC	25,3	%	30 ~ 38
RDW - CV	13,4	%	11,5 ~ 14,5
Thrombocytes	319	10 ⁹ /mm ³	150 ~ 440
Erythrocyte Sedimentation Rate	deficient	mm	0 ~ 10
Hemostasis			
PT	11,7	second	9,4 ~ 11,3
INR	1,14	-	0,83 ~ 1,10
Control	11,1	second	9,2 ~ 15,3
APTT	31,0	second	28 ~ 40
Control	32,1	second	26 ~ 37
Immunoserology			
HBsAg (ELFA)	Non Reactive	-	Non Reactive
Clinical Chemist			
Random Glucose Test	87	mg/dL	<180
Natrium (Na)	142	mmol/L	135 ~ 145
Kalium (K)	4,60	mmol/L	3,5 ~ 5,5
Chloride (Cl)	109	mmol/L	98 ~ 109
AST (SGOT)	48	U/L	0 ~ 37
ALT (SGPT)	28	U/L	0 ~ 40
Ureum	10	mg/dL	20 ~ 40
Kreatinin	0,3	mg/dL	0,8 ~ 1,5

Table 1. Result of laboratory examination.

The parents were concerned about inability of breastfeeding and the mass growth that concern getting bigger. Therefore we

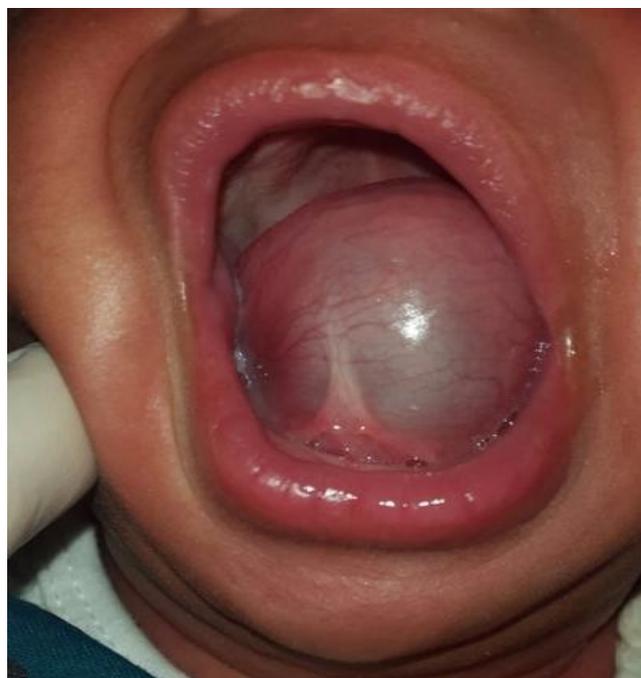


Figure 1. Patient presentation upon initial examination. Swelling on the ventral side of the tongue with the color similar with surrounding mucosa.

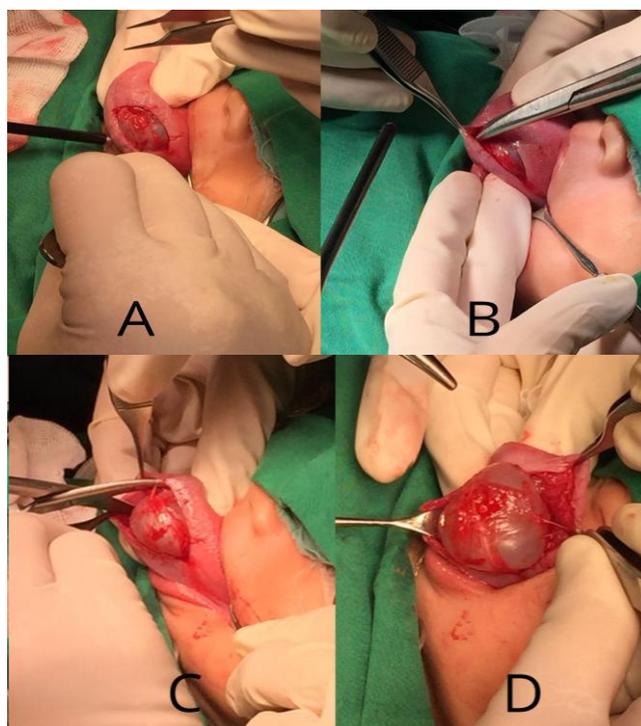


Figure 2. (A) Incision line, (B and C) Blunt dissection, (D) Mass identification.

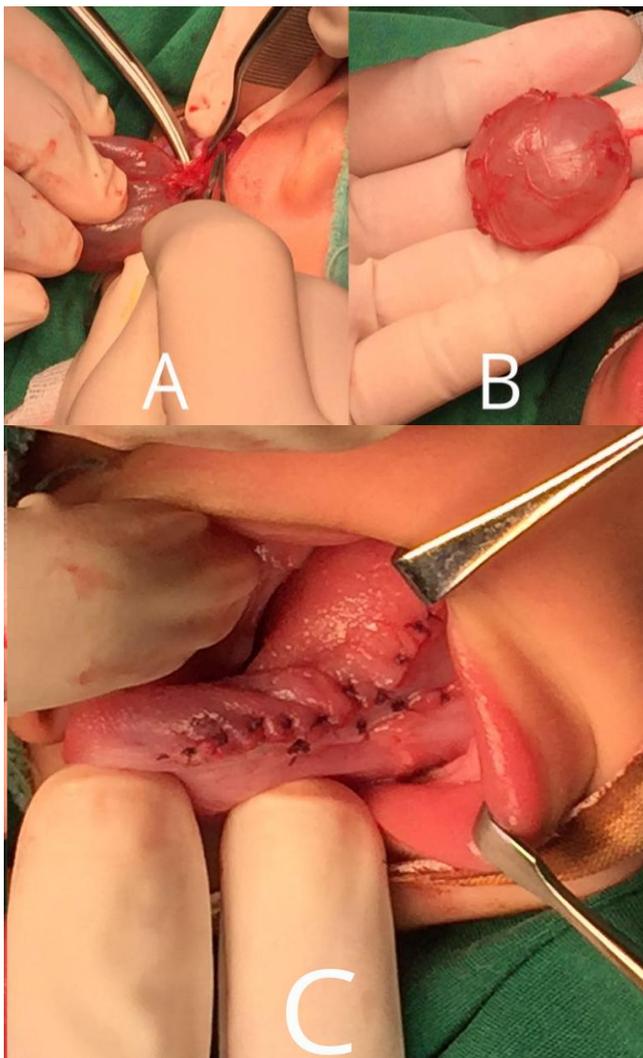


Figure 3. (A) Extirpation with clamp, (B) The mass, (C) Sutured surgery site.



Figure 4. Patient presentation upon post operation (A) Intraoral (B) Extraoral.

An intraoral approach was utilized to access the mass. Incision line was put on ventral side of the tongue mucosa with blade no. 15 (Figure 2a). Blunt dissection then performed with

blunt scissor (Figure 2b and c) and the mass was identified (Figure 2d). Continued the blunt dissection to remove the mass without breaking the mass membrane, extirpation of the mass was done by surgical clamp (Figure 3a and b). The surgery site was irrigated with NaCl 0,9% afterward sutured with Vicryl 5.0 (Figure 3c). Immediately the mass was fixated with formaldehyde buffered 10% and send for histopathological examination.

Five days after surgery, the result of histopathological analysis revealed that the mass macroscopically be in the form of white mucus mass tissue with 3 cm of measurement and microscopically consists of cyst with fibrotic tissue wall and simple columnar epithelium. This result concluded as ranula. Patient underwent uneventful recovery (Figure 4a and b). Ten months after the surgery, we followed up the patient's condition and found that there is no recurrence nor present complaint.

Discussion

This report shows a rare case of ranula which growing since the baby was born. This type of ranula can be identified as neonatal ranula and the etiology still remain unknown. Mostly ranula occurred on the floor of the mouth but in this case occurred on the ventral side of the tongue.¹ Some literatures defined the neonatal ranula as congenital ranula by reason of believed that the cause of ranula in pediatric patients might be congenital anatomic variation of the ductal system of the sublingual salivary gland.⁷⁻⁹

Proper anamnesis and physical examination are mandatory to avoid misdiagnosis. In this case, the patient's parents had chief complain of swelling on patient's tongue which slowly growing since the patient was born, also there is disturbance of breast feeding. Determining neonatal anomalies can be achieved by doing comprehensive anamnesis, especially history of the chief complain. To localize the lesion and define the cystic nature, right physical examination needs to be done. Thereupon we recorded a large cystic ranula of 3 cm in diameter with color same as surrounding mucosa in patient's ventral side of tongue. The diagnosis of neonatal ranula is concluded upon not only anamnesis and physical examination but also histopathologic examination.^{4,8}

Choristomas as differential diagnosis of neonatal ranula can be define as histologically normal tissue that is present in an abnormal location, this means the tissue would be normal if present outside of the head and neck region.⁴ The difference between ranula and epidermoid cyst is the lining epithelium, keratinized epithelium can only be found in epidermoid cyst.⁶ Other differential diagnosis of ranula is lymphangioma which histologically present thin walled, vascular channels lined with flattened epithelium.⁵ Therefore ranula can be differentiated from choristomas, epidermoid cyst, and lymphangioma histologically.

During early postnatal period, presence of intraoral cyst may cause breathing and feeding difficulty depending on the location and size of the cyst.¹⁰ In 2011, Marc Pan found intraoral cyst with symptoms of increased drooling and recent onset of snoring which exhibit signs of airway obstruction.¹¹ Larger cyst could lead to dyspnea or dysphagia and symptoms usually more acute.¹⁰ To prevent cyst in patient's tongue become larger and causing more complications, the complete extirpation was chosen as treatment plan.

As the preoperative procedure, the patient needs pre surgical preparation by pediatrician and anesthesiologist. In this case, the patient had systemic disturbances based on laboratory and radiographic examination. Limphocyte and neutrophil count showed chronic infections, we suspect this condition related to the radiographic imaging of patient's lung. However, after several test and treatment from pediatrician and anesthesiologist, the patient finally ready for surgery. We can say that multidisciplinary approach to treat pediatric patients is important to avoid surgical complication.

The surgery were performed in general anesthesia to manage infant's behavior while undergo the treatment.¹² Most ranula cases have predilection site at floor of the mouth and let us have two options to approach the lesion, intraoral and extraoral. However in this case, the lesion located in ventral side of the tongue so performing excision via intraoral approach arguably the best option.¹³ Blunt dissection then performed to release the mass from surrounding tissues without making injury in vital tissues (nerves, glands, blood vessels). Once the mass was identified, the complete extirpation of the mass with the lining membrane must be done

properly because many case reports and research on ranula show that incomplete or partial removal of cystic lining is responsible for recurrence.¹⁴ In this patient, there is no recurrence in ten months after surgery but regular check up still needed to prevent unwanted condition.

As an alternative treatment for ranula, There are 3 reported ranula cases which treated using botulinum toxin type A. Comparing to surgical treatment, injection of botulinum toxin type A (botox) has some advantages such as no need to use general anesthesia, also less injury potential to lingual nerve and submandibular duct. Botulinum toxin acts by the chemical denervation of the secretomotor parasympathetic nerve endings responsible for salivation. Its safety and tolerability are well documented.^{15,16}

Histopathological examination after surgery is mandatory to confirm the working diagnosis. In patient exhibit clinical signs of malignancy from physical examination, biopsy of the cystic wall is recommended to be done because the presence of squamous cell carcinoma arising from the cyst wall and papillary cyst adenocarcinoma of the sublingual gland may resemble ranula.¹⁷

Conclusions

Neonatal ranula is a rare case which can cause function disturbance even fatal risk. It is important to anamnesis comprehensively and perform adequate examination in order to get the right diagnosis hereupon ideal treatment plan. Noted that the neonatal cases need multidisciplinary involvement to achieve the best result.

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Declaration of Interest

The authors report no conflict of interest.

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