

(CASE REPORT)



## Sialolithiasis in children

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### Abstract

**Introduction:** Sialolithiasis is a disease in which calcified substances accumulate in the ducts or parenchyma of the salivary glands. This disease can occur in people of all ages but is mostly seen in middle-aged adults and rarely in children. This case report aimed to describe a case of sialolithiasis in children

**Case Report:** A 3-year-old girl presented with a lump that enlarged progressively on the right side of the neck since 1 year ago. The patient also complained of painful swallowing. Palpation of the right submandibular revealed a solid, mobile mass measuring 0.5x0.5cm without tenderness or hyperemia. A neck ultrasound showed an enlarged submandibular gland and right parotid gland. A midface and neck multi-slice computed tomography scan (MSCT) supported the diagnosis of sialolithiasis. The patient underwent stone removal with an extraoral approach and general anesthesia. The patient then was discharged with cefixime, methylprednisolone, and paracetamol.

**Discussion:** Sialolithiasis that cannot be removed by opening the canal can be removed by trans-oral or extra-oral sialolithotomy, extracorporeal lithotripsy, sialendoscopy, or laser treatment. Submandibular stones can be removed surgically by intraoral or extraoral approaches. Extraoral approaches are indicated for intra-glandular stones and stones embedded in the hilum of the gland.

**Conclusion:** The diagnosis of sialolithiasis in this patient was confirmed through history taking, physical examination, ultrasound, and a CT scan. Management of sialolithiasis focuses on removing the stone and maintaining salivary gland function. In this case, the stone was removed using an extraoral approach under general anesthesia which resulted in a good outcome

**Keyword:** Sialolithiasis; Children; Salivary Gland; Submandibular; Parotid

### 1. Introduction

There are three pairs of salivary glands in humans: the parotid, submandibular, and sublingual. The parotid gland consists primarily of amylase-rich saliva that secretes serous acini. The sublingual glands secrete mucus, a thick solution rich in mucin. The submandibular gland consists of a mixed population of acini with mucosal and serous functions. These three main salivary glands contribute more than 90% of saliva secretion [1].

Sialolithiasis is a disease in which calcified substances accumulate in the ducts or parenchyma of the salivary glands. This disease can occur in people of all ages but is mostly seen in middle-aged adults and rarely in children. Due to anatomical and physiological characteristics, sialoliths generally begin to form in the submandibular glands. The main symptoms of sialolithiasis are pain and swelling. Asymptomatic cases have also been reported. Clinical examination and radiological examination are used to diagnose sialolithiasis [2].

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Sialolithiasis is a common disease of the salivary glands. Symptoms include swelling of the involved gland, especially during eating, and tenderness which may subside but may recur. Sialoliths occur mainly in the submandibular glands (80-90%) and to a lesser extent in the parotid glands (5-20%). Sialoliths can be single or multiple. Multiple sialoliths in the submandibular gland are rare. Seventy out of eighty percent of cases have a single sialolith, and only about 5% of patients have three or more sialoliths [2].

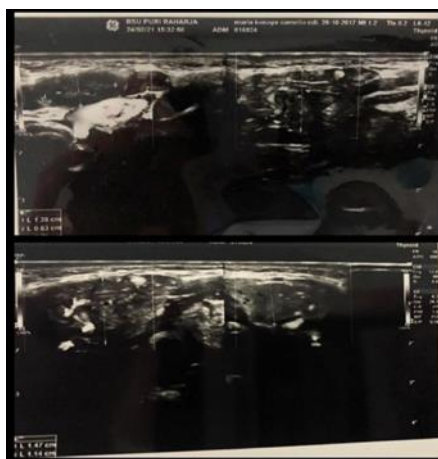
The etiopathogenesis factor associated with sialolith formation is obstruction, decreased salivary flow rate, dehydration, salivary gland infection, and impaired crystalloid solubility. Clinicians are expected to be able to diagnose early the presence of sialoliths in the salivary glands, their clinical manifestations, and management. Patients usually complain of lumps and pain when pressed, which is usually called sialadenitis. In rare cases one of the symptoms can cause objective tinnitus [3,4,5,6]. The visible difference between the submandibular and parotid glands is that the duct of the submandibular gland has a sharper angle and points upwards and the type of secretion is thicker. This makes it easier for sialoliths to form in the ducts of the submandibular glands, compared to the sublingual glands and other minor salivary glands [7].

Ultrasonography is a simple method that can demonstrate sialoliths with high accuracy. Sialoliths smaller than 3 mm can usually be detected using sialography. There are several alternatives and techniques for the treatment of sialolithiasis, including lithotripsy, sialoendoscopy, and conservative removal of the sialolith or complete removal of the submandibular gland, via transoral and extraoral routes for access to the gland. To determine the form of treatment, it is necessary to observe characteristics such as topography, diameter and location of the sialolith in the canal. Some sialoliths in salivary glands require the removal of the gland itself [7,8,9].

## 2. Case Report

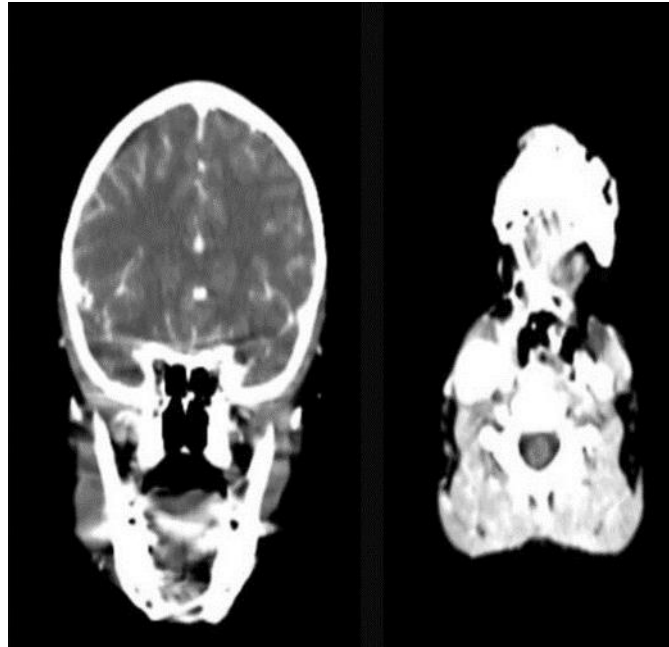
A 3-year-old girl came to the otolaryngology polyclinic at Sanglah Hospital, Denpasar, on referral from another hospital with complaints of a lump on the right side of the neck. The patient had complained of a lump on the right side of her neck since approximately 1 year ago. The lump enlarged progressively and enlarged when the patient had a fever. The patient also complained of painful swallowing. There were no coughs, colds, or shortness of breath.

On physical examination, the patient's general condition was good and alert. The blood pressure was 110/70 mmHg, pulse 80 x/minute, respiration 20 x/minute, axillary temperature 37°C. There were no abnormalities in the ears, nose, or throat. On palpation of the right submandibular, there was a solid, mobile mass measuring 0.5x0.5cm without tenderness, and no hyperemia. Complete blood tests, hemostasis physiology, and blood chemistry tests were performed with normal results. A chest x-ray examination was then carried out which showed no abnormalities. A neck ultrasound was carried out and showed an enlarged submandibular gland and right parotid gland (Figure 1). A midface and neck multi-slice computed tomography scan (MSCT) then was performed that support the diagnosis of sialolithiasis (Figure).



**Figure 1** Neck ultrasound showing mild enlargement of the submandibular gland and right parotid gland, no calcification, possible sialadenitis. Enlarged right submandibular lymph node

Based the results of the anamnesis and physical examination, as well as the supporting examinations, the patient was diagnosed with sialolithiasis. The patient underwent an extraoral approach with general anesthesia. An intravenous cefoperazone 1 gram was administered as pre-operative antibiotic therapy.



**Figure 2** MSCT Midface and axial slices without and with contrast. The calcification foci were accompanied by swelling and slight contrast enhancement in the right submandibular region, consistent with the picture of sialolithiasis with the picture of right submandibular sialadenitis.



**Figure 3** Pre-Operative Images



**Figure 4** Two-days post surgery

Based the results of the anamnesis and physical examination, as well as the supporting examinations, the patient was diagnosed with sialolithiasis. The patient underwent an extraoral approach with general anesthesia. An intravenous cefoperazone 1 gram was administered as pre-operative antibiotic therapy.

After removing the stone in the right submandibular area, a stone size 0.5 x 0.3 cm was found. The patient was advised to massage the submandibular area. The patient was discharged with cefixime 2x250mg, methylprednisolone 2x4mg, and paracetamol 3x250mg. Two days and one week follow-up revealed no complaints.



**Figure 5** One week post-surgery

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### 3. Discussion

Sialolithiasis can occur at any age, but is seen primarily in adults aged 30-60 years. This event occurs in approximately 1% of the total population, and pediatric cases are very rare, accounting for approximately 3% of all cases of sialolithiasis [2]. In this case, a 3-year-old female patient was reported with complaints of a lump on the right side of the neck accompanied by pain.

Most sialolithiasis occur in the submandibular glands, but can also occur in the parotid, sublingual, and minor salivary glands. Sialolithiasis is associated with many clinical symptoms, but the main symptoms are pain and swelling [10]. The pain in sialolithiasis is caused by the retention of saliva resulting from partial or complete obstruction of the duct by a sialolith, and subsequent expansion of the duct. These symptoms occur during or after eating [11,12]. In this case, the complaints experienced by the patient are in accordance with the literature.

Sialolithiasis is diagnosed based on radiographs and clinical examination, including medical history, inspection, and bimanual palpation of the affected salivary glands. Sialography is useful in detecting sialoliths, and high-frequency ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), and sialendoscopy can be used to diagnose sialolithiasis. Bimanual palpation of the oral cavity is useful for examining sialoliths in the submandibular gland ducts [13]. The patient underwent a neck ultrasound and a CT scan with the impression of sialolithiasis.

In this case, the stone is visible from the outside so the patient is treated surgically using an extraoral approach and antibiotics are given. Treatment is determined by the size, location, amount of sialolith, and the degree of interference with excretion from the ducts. Conservative treatments, such as hydration, massage of the affected salivary glands, warming, and administration of sialagogues, antibiotics, and non-steroidal anti-inflammatory drugs (NSAIDs), may be used in cases where the sialolith is smaller than the diameter of the duct [2]. Sialoliths that cannot be removed by opening the canal can be removed by trans-oral or extra-oral sialolithotomy, extracorporeal lithotripsy, sialendoscopy, or laser treatment [8,14]. Submandibular stones can be removed surgically by intraoral or extraoral approaches. Extraoral approaches are indicated for intra-glandular stones and stones embedded in the hilum of the gland. This procedure shows satisfactory clinical results, and is simple to perform by oral and maxillofacial surgeons [15]. Other studies suggest that extraoral approaches to remove submandibular glands are relatively simple and widely accepted. However, the extraoral approach poses a risk to the marginal mandibular nerve [16].

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#### 4. Conclusion

Sialolithiasis in children is a rare case. A case has been reported in a 3-year-old girl. The diagnosis of sialolithiasis in this patient was confirmed through history taking, physical examination, and supporting examinations. In this case, the supporting examination chosen was an ultrasound and a CT scan. Management of sialolithiasis focuses on removing the stone and maintaining salivary gland function. There are various techniques for removing salivary gland stones. In this case, the stone was removed using an extraoral approach under general anesthesia which resulted in a good outcome.

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#### Compliance with ethical standards

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##### *Disclosure of conflict of interest*

There are no conflicts of interest.

##### *Statement of informed consent*

Informed consent was obtained from participant included in the study.

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