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# Bronchogenic CYST of the tongue in child diagnostic difficulties and role of surgery in the treatment (about 2 cases)

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**Abstract**
Our study aims to show essentially the difficulties of diagnostic of bronchogenic cyst of the tongue in child, as well as the place of surgery in the diagnosis and treatment of these cystic masses.
We report the cases of two children with bronchogenic cyst, treated in the Pediatric Surgery Department of the Children's Hospital in Rabat.
A 6-month-old girl and a newborn at j 5, both admitted for feeding difficulties. The clinical exam of the tongue finds a cystic lesion. The imaging examination demonstrated the fluid nature of the mass, and allowed to analyze the reports of the cyst. The patients were operated. The postoperative follow up was simple. The long-term evolution was satisfactory, with no sign of recurrence or complication.
Histological examination concluded on bronchogenic cyst characterized, showing respiratory epithelium.
The absence of any clinical or radiological specificity of the bronchogenic cyst, only histology can certify the diagnosis, based on the demonstration of a respiratory epithelium associated with elements of the tracheal bronchus tracts.
The treatment of the bronchogenic cyst is surgical. The prognosis is mainly conditioned by the quality of the excision, which must be complete.
The Immediat and long term follow up must always be in place.

**Keywords:** bronchogenic cyst, tongue, surgery

## Introduction
The bronchogenic cyst is a choristoma affecting the upper aerodigestive tract; it is lined with a respiratory epithelium.
It usually sits at the level of the mediastinum and more rarely at the level of the pulmonary parenchyma.
Other localizations have been described, in particular intra - pericardial, abdominal or cutaneous.
The ectopic bronchogenic cysts of the tongue, which will be our study, are extremely rare, with less than ten cases described until today in the literature.

## Materiels and Methods
We report in our study two cases were followed in Pediatric Surgery department in children 's hospital for bronchogenic cysts of tongue.
A 6-month-old girl and a newborn at j 5, both admitted for feeding difficulties.
The treatment in these cases is surgical. The anatomopathological study confirmed the diagnosis.

## Results
### Case 1
Girl had 6 months old years, without particular pathological antecedents. No notion of consanguinity in the parent.
At the birth, the mother noted a swelling at the level of the tongue gradually increasing volume, causing disorders of suction and swallowing.

### Examination on admission
The inspection: found a swelling occupying almost the entire lower surface of the tongue (2/3 of the ventral side) causing a protrusion there of outside the oral cavity which remains open (the tip of the tongue is elevated up and forward). Figure 1

**Fig 1:** lingual mass

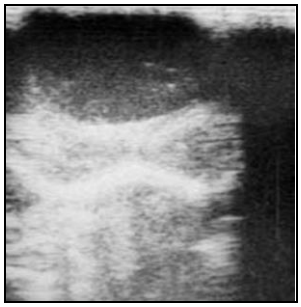

**Fig 2:** ultrasound of the tongue showing

The palpation: note a mass of liquid consistency, immobile, renitent, measuring approximately 2X2 cm without associated inflammatory signs.

**Examination of ganglionic areas was normal**
The rest of the somatic examination is without particularity; without associated malformations.

**Para-clinical examinations**
Ultrasound of the tongue: confirms the cystic character of the mass, it is delimited by a wall. (Figure 2)
CT: shows a process of lesionnel of the mobile segment of the tongue, hypodense, not raised by the intravenous injection of the product of contrast, well limited by a dense and thin wall. This medial process deforms the external contours of the tongue and measures 23 x 21.5 x 21mm with no cervical lymphadenopathy and associated bone lesions.
Chest X-ray: normal Abdominal ultrasound: without particularity.
On all of these clinical and para-clinical data, two diagnoses have been mentioned: the dermoid cyst and digestive duplication.

**Treatment The indication for surgery was chosen**
The child underwent general anesthesia after nasotracheal intubation, and traction of the tip of the tongue outside the mouth, a T-shaped incision on the ventral side of the tongue is performed. The dissection of the mass with respect to the mucosa and to the different lingual muscles allowed its liberation and total excision.

**The gastric tube is kept 48 hours postoperatively**
Histological examination of the surgical specimen confirms the diagnosis of the bronchogenic cyst by showing a cystic formation whose wall is lined by a pseudo-stratified cylindrical epithelium of the respiratory type with a focus of regular squamous metaplasia. The underlying chorion is fibrous and rich in lymphocytes with no other heterologous tissue structures. This cyst is surrounded by a band of striped muscular tissue of the lingual type. Figure 3

**Fig 3:** Histological section showing cylindrical and ciliated pseudo-stratified respiratory epithelium.

The operative follow-up was simple except for a small edema of the rapidly resolving tongue under medical treatment, the oral feeding was resumed on the 3rd day without difficulties and the child was released on the 5th day.
Long-term evolution: The child is seen again five months later in consultation, the tongue is completely healed and the somatic examination is completely normal.

**Case 2**
Identity: newborn male at J5 admitted for mass of the tongue.
Without particular pathological antecedents.
At the birth, the parents note a voluminous mass of the tongue preventing the closing of the mouth and hindering the feeding.
Examination of the tongue fuond a bulky cystic mass.
Localized in the tip of the tongue and the sublingual area, measuring approximately 3 x 4cm, preventing oral closure, without associated inflammatory sign.
Examination of ganglionic areas was normal.
The rest of the somatic examination is without particularity.
Any associated malformations are noted.
Ultrasound of the tongue: confirms the cystic character of the mass.
It is delimited by a clean wall and regular contours.
CT noted a mass of hydrous density, rounded, well limited by a thick and regular wall not raised by the product of contrast, including neither calcifications nor partitions.
Treatment: The indication for surgery was chosen.
A puncture of the cyst showed a yellowish liquid with viscous aspect. The tumor was resected by partial glossectomy. After incision in inverted "V" on the tip of the tongue The reconstruction was ensured by simple sutures in 3 planes.
Histological examination confirmed the diagnostic of bronchogenic cyst.

**Discussion**
The bronchogenic cyst is a rare congenital malformation. It is part of the choristomes, that is benign embryonic tumors characterized by the presence in an abnormal location of normal tissue (heterotopy).
Bronchogenic cysts are located in 84% of endothoracic cases, particularly at the mediastinal level, thus representing 10% of the mediastinal masses of the child [1, 2].
The cysts are of ectopic localization in 16% of cases, which can be found at the cervical level, the esophageal or thoracic wall, the skin, the pericardium, the diaphragm and the abdomen.
Lingual KBs are rare. In a review of the literature made between 1942 and 1997, Manor *et al*. Described 52 lingual cysts, considered to be bronchogenic or digestive choristomas [3].
Of the 7 cases described in the literature, 6 were male (85.71%) and 1 female (14.29%). There is no explanation for this male predominance The age of diagnosis depends mainly on the volume of the cyst and the severity of the clinical picture.

**1. Diagnostics**
**a. Antenatal diagnosis**
The antenatal diagnosis of the bronchogenic cyst of the tongue can be done with obstetric ultrasound performed in a systematic way, it makes it precise topographic and structural description and strives, especially to verify the entirety of the organs [4].
The fetal ultrasound present an intra-lingual unilocular cystic formation, in the third trimester [5, 6]. ; it evokes the bronchogenic cyst of the tongue.
MRI, a powerful examination, may be of great interest in the diagnosis of lingual abnormalities, and may complement antenatal ultrasound to clarify the lingual origin of an abnormal mouth. In antenatal, MRI can show the bronchogenic cyst as a mass with hyper signal in T2, which

is invisible in T1 [7].
In the end, we highlight the value of antenatal diagnosis.
Thus any lingual malformation detected before birth requires the immediate and adapted care of the newborn by a specialized multidisciplinary team.

**b. Clinical**
There is no clinical specificity of the lingual bronchogenic [8, 9].
The symptomatology will therefore depend essentially on the volume of the cyst: As this one is very variable, the polymorphism of the symptoms is very important. We will note :
- Food difficulties, sucking, chewing and swallowing disorders.
- Disorders of phonation and speech.
- Difficulties learning.
- Macroglossia preventing mouth closure.
- Nocturnal vomiting has also been reported once in the literature [3].
The clinical examination shows during the inspection a lingual mass in the form of a submucosal bulge easily recognizable as in our observation.
The seat of the mass at the level of the tongue is variable which may interest, the posterior or anterior side, the movable or fixed part.
Its volume also differs, with extremes according to the literature of 1 to 6 cm of diameter at the time of the consultation.
This is usually a single cyst, as is the case with our two patients.
The bronchogenic cyst of the tongue is yellow or yellow-white, negative for transillumination [10].

**c. Paraclinic**
Ultrasound is the first-line examination that confirms the cystic nature of the lesion.
It allows to specify the characteristics of the mass to know:
The bronchogenic cyst generally corresponds to a hypoeechogenic and homogeneous rounded image with regular edges delimited by a clean wall.
Ultrasound has its limits especially in the precision of the extension in depth [11].
It was performed in first intention in both our patients.
CT have a role both in the positive diagnosis and in the accuracy of the lesional extension by appreciating the seat, the topography, the depth of the mass as well as the associated bone lesions.
The acquisitions in helical mode and in particular with the multiple detectors allow a volume acquisition and reconstructions in the three planes of the space equivalent to those provided by the MRI.
The mass appears in the form of a well-defined lesion with a thin or thick wall, a fluid density ranging from -10 to +10 HU, and which does not take on contrast.
The occurrence of infectious or haemorrhagic complications leads to a changé in its size but also an increase in intralesional density, or even a parietal enhancement [12].
MRI is a powerful technique in the study of abnormalities of the base of the tongue, given its superiority in soft tissue analysis and in the distinction between muscles and pathological masses. The exploration is carried out in the different planes of the space.
It usually finds a well-limited mass rounded frequently in hypersignal in T2.
This hypersignal is probably explained by the presence of protein material in these cysts [12, 13].

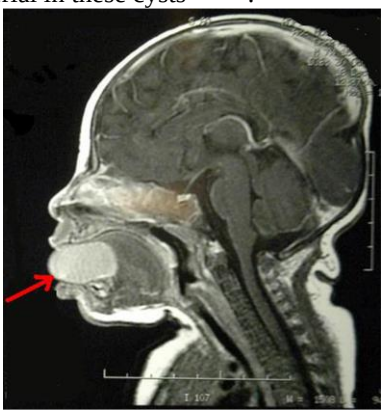

**Fig 4 :** Sagittal section of MRI in T1 with gadolinium injection showing a lingual cystic image [14].

Standard radiography: It must be made in front and side incidence, looking for other localizations: mediastinal and pulmonary.

**d. Histology**
The diagnosis of certainty is made on the surgical excision specimen because only the anatomopathological analysis and the immunohistochemistry can affirm the respiratory nature of the epithelium lining the cyst [15].

**Macroscopy**
Macroscopically it is about rounded tumors, of gray pink color, of variable volume, often smaller than the radiological aspect suggests.
Their wall is thick, smooth, well vascularized and surrounded by a strong shell. The cystic pouch is single or multiple.
The cysts are filled with thick, viscous mucus secreted by the bronchial-type gland cells contained in the chorion [16]. he chest x-ray in both patients was normal.

**Microscopy**
Bronchogenic cysts are classified as dysembryoplasias also called hamartomas. It is a tumor-like tissue malformation composed of an abnormal mixture of constitutive elements normally present in the thoracic cavity.
Histology provides evidence of the bronchogenic origin of the cyst by finding a ciliated epithelium pseudostratified respiratory type.
This epithelium consists of tall cylindrical cells with a basal nucleus. The upper pole is provided with eyelashes that float in the cyst lumen. These cells secrete mucus that accumulates in the cystic cavity.
Areas of squamous metaplasia can be found in places. The wall of bronchogenic cysts may also contain glandular, cartilaginous and smooth muscle elements. Their presence unambiguously signifies the bronchogenic origin of the cyst [13].
The anatomopathological study of the excision specimen was performed for our two patients with a result in favor of a bronchogenic cyst of the tongue.

**2. Treatment**
**a. Therapeutic means**
**Cystectomy**

It consists of a complete tumor excision while preserving as much as possible the integrity of the different muscles of the tongue and sublingual salivary glands [16].
Surgery is performed under general anesthesia with nasotracheal intubation.
After performing a perfect brushing of the oral cavity, we will proceed to pull the tongue out of the mouth (up and forward) with a wire placed at the tip of the tongue.
The most convenient approach is a horizontal or longitudinal incision next to the cyst.
The dissection of the cyst will be done with electrocautery and / or chisel, leaving the lingual muscles and above all the sublingual salivary glands, which can have intimate relations with the wall of the cyst [17].
Once the cleavage plane is found, the cyst can be removed without any problem.
After performing a perfect hemostasis, we will close the incision by suturing the banks.
We will finally perform oral hygiene with betadine serum.
The nasogastric feeding tube can be kept for a few days pending the resolution of peri-lesional edema.
n our study, the cysts were located at the level of the mobile portion of the tongue, the ventral side in the girl and the tip in the boy, without any contact with the salivary glands.
For both patients, the procedure was performed under general anesthesia with naso tracheal intubation. The excision of the cyst proceeded according to the steps described above without any intraoperative complication.

**Cystic puncture**
It is an aspiration puncture that precedes the cystectomy, usually when the cyst is very large.
The puncture can also sometimes guide the diagnosis but is possible, easily, only in the adult, that is to say in very few cases. It was performed at our second patient, under general anesthesia at the time of the operation, given the huge volume of the cyst. She brought back a yellow liquid of viscous appearance.

**b. Indications**
The operative indication is formal because of the risk of complications such as:
-the infection, haemorrhage the rapid increase in the volume of the cyst may cause respiratory, nutritional or phonetic disorders.
When the cyst is large, a previous aspiration puncture may be useful, facilitating its removal, as was done in our second observation [16].
Surgical enucleation of the lesion can be performed for diagnostic and therapeutic purposes and for the prevention of possible complications.
It must take place according to several studies as soon as the first complications occur or around the age of one to make a compromise between the risks linked to general anesthesia, the latter decreasing with age, and the risks associated with possible complications [14].

**c. The operative follow-ups**
They are usually simple.
Complications can occur such as bleeding, infection, edema and fever. However, no postoperative complication has been reported to date in the world literature.
In our two observations, the operative follow-ups were also simple, apart from a small lingual edema rapidly resolving under medical treatment in our patient.

**d. The evolution**
The prognosis is mainly conditioned by the quality of the excision, which must be complete.
In this case, the evolution is generally favorable.
However, short- and medium-term surveillance should be used, especially if there are dysplasia areas.
The risk of malignant degeneration to bronchoalveolar carcinoma, adenocarcinoma or squamous cell carcinoma is still possible, although rarely reported in the literature. This risk is even lower if the bronchogenic cyst is treated early and in a radical way [16].
Our first patient was reviewed in consultation 5 months later. His somatic examination was perfectly normal with no sign of recurrence or complication.

**Conclusion**
In the majority of cases, bronchogenic cysts are diagnosed and treated in childhood.
The symptomatology is no specific, very variable depending on the volume of the cyst, ranging from simple discomfort during feeding to a true asphyxic table.
The diagnosis of certainty is histological, finding a respiratory epithelium; pseudo-stratified, associated with elements of the tracheobronchial Surgical excision is the definitive treatment of the lesion. It is a complete excision of the mass respecting the neighborhood structures.
The evolution is generally favorable if the excision is complete. The recurrences are exceptional and no case has been described until today.

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