

Systematic Review

Hydatid Cysts of the Tongue: A Systematic Review of the Literature

Nawshad Muhammed¹, Gulmina Saeed Orakzai², Shamsul Hadi³, Usman Ashraf⁴, Tri Shaikh⁵, Abul Khair Zalan⁶, Vishal Lohana⁷, Salman Shams⁷, Sumia Rami⁷, Hamna Hoor^{8*}

1. Department of Dental Materials, Institute of Basic Medical Sciences, Khyber Medical University, Peshawar, Khyber Pakhtunkhwa, Pakistan
2. Watim Medical and Dental College, Islamabad, Pakistan
3. Department of Pathology, Saidu Group of Teaching Hospital (SGTH), Swat, Khyber Pakhtunkhwa, Pakistan
4. Department of Orthodontics, University College of Medicine and Dentistry, The University of Lahore, Lahore, Punjab, Pakistan
5. Oral & Maxillofacial Department, Isra Dental College, Isra University, Hyderabad, Sindh, Pakistan
6. Department of Pediatric Dentistry, School of Dentistry, PIMS, Islamabad, Pakistan
7. Oral Medicine Department, Liaquat University of Medical and Health Sciences Jamshoro, Sindh, Pakistan
8. University College of Medicine and Dentistry, The University of Lahore, Lahore, Punjab, Pakistan

* Corresponding author: hamnahoor83@gmail.com (H. Hoor). 73-C, DHA Rahbar, Zip code: 2023, Lahore, Pakistan



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Abstract

Introduction

Hydatid cysts, caused by *Echinococcus granulosus*, typically affect the liver and lungs but can rarely occur in the tongue, hence presenting as a diagnostic challenge

Methods

This is a systematic review of the reports of hydatid cysts in the tongue up to April 2024, emphasizing diverse clinical presentations, diagnostic complexities, treatment modalities, and outcomes.

Results

Six cases of tongue hydatid cysts were reported in the literature, of whom three were male, two were female, and the gender of one patient was not determined. The patients were between 3 and 35 years old, with a mean age of 15 years. Only one case has hydatid cysts in the other organs, namely the liver and lung. Four cases were reported in developing countries. Clinical presentations varied, including tongue swelling, speech articulation difficulties, and swallowing impairments.

Conclusion

Hydatid cyst of the tongue has a possible endemic nature. It usually presents with painless tongue swelling. The primary management strategy is surgical excision, which has a favorable outcome.

1. Introduction

Hydatid cysts result from *Echinococcus granulosus* infection [1]. The parasite has a life cycle involving canids as definitive hosts and herbivorous mammals as intermediate hosts. Canids

ingest parasite eggs, which hatch into larvae in their intestines. These larvae form hydatid cysts in the organs of intermediate hosts. When canids consume infected tissues, cysts release larvae that mature into adult tapeworms in the intestine, completing the cycle [2]. Hydatid cysts usually affect the liver and lungs; infrequently, they manifest in extrahepatic-extrapulmonary sites, such as the diaphragm, chest wall, breast, musculoskeletal system, intestine, Kidney, bladder, heart, brain, and abdominal wall [3-12]. Neck involvement has rarely been reported in developing countries [13-16]. Tongue involvement poses unique diagnostic challenges due to its rarity and nonspecific clinical features. This review analyzes the reported cases of hydatid cysts in the tongue, elucidating the diverse clinical presentations, diagnostic approaches, treatment strategies, and outcomes, aiming to enhance understanding of this rare entity.

2. Methods

A systematic literature search was conducted to identify case reports documenting hydatid cysts involving the tongue. Inclusion criteria encompassed cases reported until April 2024, with confirmed diagnosis through clinical and histopathological examination. The references of the included articles were also examined for papers reporting hydatid cysts of the tongue. Data extraction included patient demographics, presenting symptoms, diagnostic methods, surgical techniques, histopathological findings, and postoperative outcomes. Studies published in predatory journals were excluded from the study. As the number of cases expected to be minor, incomplete articles and non-English reports were not excluded from the review.

3. Results

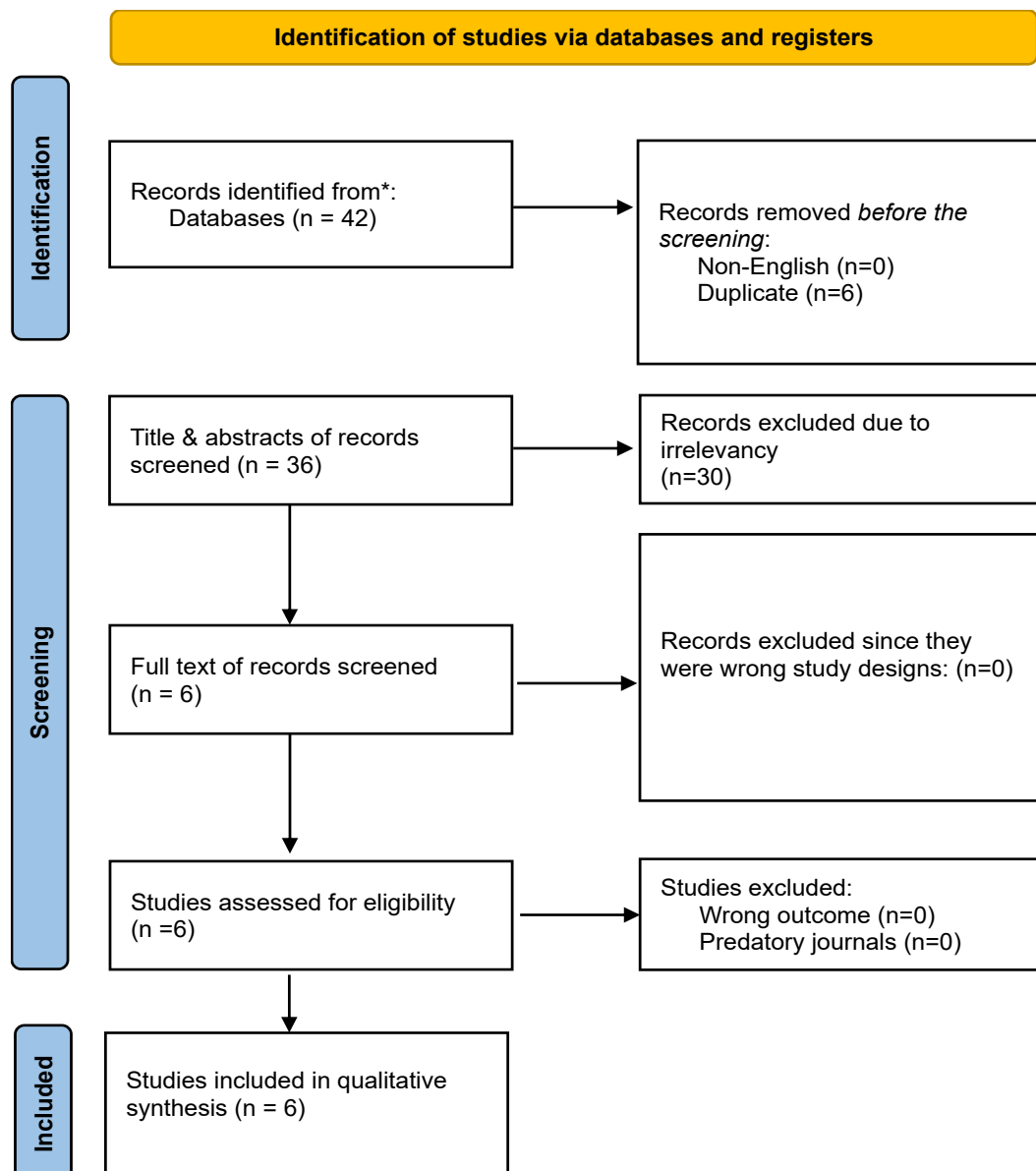


Figure 1. Study selection PRISMA flow chart

Table 1. Review of the reported cases of hydatid cysts of the tongue.

| Author/Year | Country | Age (year)/Sex | Presentation | Diagnosis | Management | Hydatid cysts of other organs |
|---------------------------|--------------|----------------|--|--|-----------------------------------|-----------------------------------|
| Aggarwal et al. 2017 [17] | India | 10/male | Tongue Swelling, difficulty in swallowing | Histopathological examination of the cyst wall | Spontaneous rupture and expulsion | None |
| Perl et al. 1972 [1] | South Africa | 18/female | Tongue swelling | Growth examination of the specimen | Excision | None |
| Saez et al. 2001 [19] | Chile | 3/female | Tongue swelling | Histopathological examination of the specimen | Excision and antihelminth | Involvement of the liver and lung |
| Gracanin 1963 [20] | Yugoslavian | 35/male | Tongue swelling, difficulties in speaking, swallowing, and mastication | --- | --- | --- |
| Goel et a. 1974 [21] | India | --- | --- | --- | --- | --- |
| Tahiri 1965 [22] | --- | 9/male | --- | --- | --- | --- |

* missing data are denoted by ---.

The review identified six distinct cases of hydatid cysts (Figure 1), of whom three (50%) were male, two (33.3%) were female, and the gender of one patient (16.7%) was not determined. The patients were between 3 and 35 years old, with a mean age of 15 years. Only one case has hydatid cysts in the other organs, namely the liver and lung. Most cases were reported in developing countries (Table 1). Clinical presentations varied, including tongue swelling, speech articulation difficulties, and swallowing impairments. Diagnostic evaluation primarily did

not rely on imaging modalities, such as computed tomography (CT) scan, magnetic resonance imaging (MRI), and ultrasonography, while confirmed by histopathological examination following surgical excision. Surgical intervention, aiming for complete cyst removal while preventing rupture, was the cornerstone of treatment, yielding favorable outcomes with no recurrence observed during follow-up assessments.

4. Discussion

The incidence of hydatid cysts spans a wide age range, reflecting the diverse demographics of affected individuals. While cases occur across all age groups, pediatric populations exhibit a notable presence, highlighting the indiscriminate nature of this parasitic infection. Children may acquire the infection through various means, including contact with infected animals or ingesting contaminated food or water. Additionally, the prevalence of hydatid cysts in endemic regions contributes to the higher incidence observed in younger age groups. In contrast, cases in older individuals may present distinct challenges, given

the potential impact of age-related comorbidities on diagnosis and treatment outcomes. Understanding the age distribution of hydatid cysts is essential for clinicians to recognize and manage this condition effectively across different age demographics, ensuring timely intervention and optimal patient care [23]. The patients diagnosed with hydatid cysts of the tongue, as documented across the reported cases, exhibit a diverse age distribution. While most cases fall within the pediatric to young adult age range, ranging from 3 to 35 years, a case involving a 3-year-old patient notably contributes to this spectrum. Consequently, the average age of patients affected by hydatid cysts of the tongue stands at approximately 15 years. This broader age range underscores the importance of maintaining a high index of suspicion for hydatid cysts across pediatric, adolescent, and adult populations presenting with tongue swellings, facilitating timely diagnosis and appropriate management tailored to different age groups [17-22].

The clinical presentation of hydatid cysts, in general, varies depending on the location and size of the cyst, as well as the affected organ. Common manifestations include asymptomatic cysts detected incidentally during imaging studies or symptomatic cysts causing localized or systemic symptoms. In the liver, the most common site of hydatid cysts, patients may present with abdominal pain, hepatomegaly, jaundice, or signs of biliary obstruction. Pulmonary hydatid cysts can manifest with cough, chest pain, dyspnea, or hemoptysis. Hydatid cysts in other organs, such as the brain, spleen, or bones, may present with specific neurological deficits, abdominal pain, or localized swelling, respectively. Complications such as cyst rupture, anaphylaxis, or secondary infection can occur, leading to acute exacerbation of symptoms [24-28]. The presentation of hydatid cysts affecting the tongue is characterized by a spectrum of clinical manifestations, often posing diagnostic challenges due to its rarity and nonspecific symptoms. Patients typically present

with a gradually enlarging tongue swelling, which may be accompanied by discomfort, difficulty in speech articulation, mastication, and swallowing. The clinical course may vary, with some patients experiencing intermittent pain or exacerbation of symptoms over time. Additionally, a history of contact with dogs, residing in endemic regions, or engaging in activities associated with parasitic transmission may provide valuable clues to the etiology. Differential diagnoses may include benign and malignant lesions, necessitating thorough evaluation through imaging studies [17-22].

The diagnosis of hydatid cysts relies on a combination of clinical evaluation, imaging studies, serological tests, and histopathological examination. Imaging modalities such as ultrasound, CT, and MRI play a central role in visualizing cystic lesions and assessing their size, location, and relationship with surrounding structures. In soft tissue involvement, ultrasound is often the initial imaging modality of choice due to its accessibility, cost-effectiveness, and ability to differentiate cystic from solid lesions. CT and MRI offer higher resolution and provide detailed anatomical information, aiding in surgical planning and evaluation of cyst complications. Serological tests, including enzyme-linked immunosorbent assay (ELISA) for echinococcal antibodies and antigen detection assays, can support the diagnosis, especially in cases with atypical imaging findings or inconclusive results. However, serological tests may yield false-negative results in certain circumstances, limiting their diagnostic utility. Histopathological examination of cyst fluid or tissue obtained during surgical excision remains the gold standard for definitive diagnosis, confirming the presence of characteristic structures such as protoscoleces, laminated membranes, and daughter cysts [29, 30]. Diagnosis of tongue cysts often presents challenges due to their uncommon occurrence and diverse clinical presentations. In the described cases, diagnostic methodologies varied, reflecting the complexity of identifying such lesions. Histopathological examination of specimens provided definitive confirmation in some cases, highlighting the characteristic features of hydatid cysts under microscopic scrutiny. Conversely, other cases relied on growth examination, suggesting a reliance on observing the development of the cyst within the specimen [17-19].

The management of hydatid cysts involves a multifaceted approach tailored to the individual patient's condition, cyst characteristics, and location. Surgery remains the cornerstone of treatment for most cases, aiming for complete cyst removal while minimizing the risk of cyst rupture and subsequent anaphylactic reactions. Various surgical techniques may be employed, including pericystectomy, cystotomy with evacuation of cyst contents, and capitonnage, depending on cyst size, location, and involvement of vital structures [31]. Additionally, preoperative measures such as albendazole therapy may be administered to reduce the risk of intraoperative dissemination of viable parasite material. In cases where surgery is not feasible due to cyst location or patient comorbidities, alternative interventions such as percutaneous aspiration, injection of scolical agents, and percutaneous drainage under radiological guidance may be considered, often in conjunction with adjunctive medical therapy. Postoperative management typically involves continued administration of albendazole to prevent recurrence and monitor for any residual or recurrent

disease. Long-term follow-up is essential to detect any recurrence or complications, such as secondary bacterial infection or anaphylaxis, and to optimize patient outcomes [32, 33].

5. Conclusion

Hydatid cyst of the tongue is a rare disease; it is possible in an endemic region. It usually presents with painless tongue swelling. The primary management strategy is surgical excision, which has a favorable outcome.

Declarations

Conflicts of interest: The author(s) have no conflicts of interest to disclose.

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Authors' contributions: HH was a major contributor to the conception of the study and the literature search for related studies. NM, GSO, and SH were involved in the literature review, manuscript writing, and data analysis and interpretation. UA, TS, AKZ and VL Literature review, final approval of the manuscript, and processing of the tables. SS and SR were involved in the literature review, the study's design, and the manuscript's critical revision. HH and NM Confirmation of the authenticity of all the raw data All authors approved the final version of the manuscript.

Use of AI: AI was not used in the drafting of the manuscript, the production of graphical elements, or the collection and analysis of data.

Data availability statement: Note applicable.

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