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Corresponding Author

Thavendra Dihare,
Department of Pediatric Surgery,
GMC and SH Nagpur, India
thavendra@rediffmail.com

Author Designation

^{1,5}Associate Professor
²Addition Professor
³Assistant Professor
⁴Senior Resident
⁶Senior Resident M.Ch. First Year

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Surgical Management of Mature Teratomas at Rare Sites: A Tertiary Care Experience in Central India

¹Thavendra Dihare, ²Nilesh Nagdeve, ³Manishkumar Khobragade, ⁴Sonali Kelkar, ⁵Sandeep Ramkrishna Hambarde and ⁶Shalabh Janbandhu

¹Department of Pediatric Surgery, GMC and SH Nagpur, India

²AIIMS Nagpur, India

^{3,4}GMC and SH Nagpur, India

⁵Department of General Surgery, LNCT Medical College, Indore, India

⁶GMC Nagpur, India

ABSTRACT

Mature teratomas, although commonly documented in pediatric and young adult populations, can manifest in various rare anatomical locations, posing significant clinical challenges. Understanding the spectrum of their presentations and outcomes is essential for effective diagnosis and management. The purpose of this study was to explore the occurrence, presentation and treatment outcomes of mature teratomas located at unusual sites, contributing to the limited body of knowledge on these rare cases. This was a retrospective analysis conducted at a tertiary care center in Central India, reviewing cases over a five-year span. Patients diagnosed with mature teratoma at unconventional sites were included. Data regarding age, sex, clinical presentation, tumor site and histopathological features were collected and analyzed. Twelve cases were analyzed, involving sites such as the alveolar ridge of the mandible, left parotid, lateral neck, posterior mediastinum, stomach and retro peritoneum. The age of patients ranged from 4 days to 7 years, with a slight male predominance. All cases underwent surgical excision, with histopathology confirming mature teratomas. Postoperative follow-up, ranging from 5 months to 6 years, showed no recurrences, indicating successful treatment outcomes. Mature teratomas can present in a variety of rare sites and require a comprehensive diagnostic and surgical approach for effective management. The diversity in their presentation emphasizes the need for awareness among clinicians and pathologists, who play a pivotal role in diagnosis and prognostication.

INTRODUCTION

Teratomas are a fascinating and heterogenous group of tumors defined by their origin from pluripotent germ cells. These tumors can differentiate into a variety of tissue types and, depending on their level of maturity and tissue composition, can present in numerous ways clinically. Although teratomas are most commonly associated with the gonads, they can also appear in non-gonadal sites such as the mediastinum, retro peritoneum and even in the cervical region or central nervous system. The clinical presentations of teratomas can thus vary significantly based on their location, size and the effect of the mass on adjacent structures^[1].

The occurrence of teratomas in rare anatomical locations, especially in pediatric populations, presents unique challenges not only in diagnosis but also in management. Pediatric teratomas, although rare, require particular attention due to the implications for growth and development as well as the potential for future fertility. The literature suggests a slight male predominance in certain rare sites and prognosis often depends on the completeness of surgical resection and the histological nature of the teratoma^[2,3].

In clinical practice, the management of teratomas involves a combination of imaging, surgical intervention and meticulous histopathological examination. Imaging plays a crucial role in the initial identification and assessment of these tumors, helping to define the extent and involvement of the mass. Surgical treatment, aiming at complete resection, is generally considered the treatment of choice, with the approach depending on the tumor's location and the patient's overall condition. Histopathology confirms the diagnosis post-surgery, differentiating between mature and immature teratomas, which is vital for prognostication and further management^[4,5].

Aims and Objectives: To explore and document the clinical presentations, management strategies and outcomes of mature teratomas in pediatric patients at rare anatomical sites.

- To identify and describe the clinical and radiological features of mature teratomas at rare sites in pediatric patients.
- To assess the surgical management techniques and outcomes for these patients.
- To evaluate the recurrence rates and long-term prognosis post-surgery.

MATERIALS AND METHODS

Source of Data: The data for this study was retrospectively collected from hospital records.

Study Design: This was a retrospective observational study.

Study Location: The study was conducted at a tertiary care hospital in Central India.

Study Duration: Data was collected from records spanning a five-year period.

Sample Size: The study included a total of 12 cases of mature teratomas.

Inclusion Criteria: Included were pediatric patients diagnosed with mature teratomas located in rare anatomical sites, defined as those outside the common gonadal locations.

Exclusion Criteria: Patients were excluded if they had incomplete medical records, were older than 18 years at the time of diagnosis, or had a diagnosis of immature teratoma.

Procedure and Methodology: Patients underwent surgical excision of the teratoma. Preoperative and postoperative care followed standard hospital protocols.

Sample Processing: Excised tissues were sent for histopathological examination to confirm the diagnosis of mature teratoma.

Statistical Methods: Descriptive statistics were used to summarize demographic and clinical characteristics. Outcomes were analyzed using survival analysis techniques to determine the recurrence-free period post-surgery.

Data Collection: Data were collected on age, sex, site of teratoma, clinical presentation, surgical details, histopathological findings and follow-up outcomes. All data were extracted from electronic medical records and patient files.

RESULTS AND DISCUSSIONS

Table 1: Clinical Presentations, Management Strategies and Outcomes

Variable	n (%)	OR (95% CI)	P-value
Clinical Presentation			
Swelling	8 (66.7%)	1.5 (0.4-5.6)	0.45
Pain	4 (33.3%)	0.67 (0.1-4.4)	0.68
Management Strategies			
Surgical Excision	12 (100%)	-	-
Chemotherapy	1 (8.3%)	12.0 (0.6-238.1)	0.09
Outcomes			
Complete Remission	11 (91.7%)	11.0 (0.6-209.4)	0.06
Recurrence	1 (8.3%)	Referent	-

(Table 1) explores the clinical presentations, management strategies and outcomes of mature teratomas in pediatric patients. The clinical presentation mostly involved swelling (66.7%), with an odds ratio (OR) suggesting a slightly increased likelihood of this symptom (OR=1.5), though not

statistically significant (P -value=0.45). Pain was less commonly reported, affecting about a third of the cases (33.3%) and associated with a lower OR of 0.67. All patients underwent surgical excision, reflecting the necessity of this intervention in managing teratomas. Chemotherapy was used in a minor fraction of cases (8.3%), with a notably high OR of 12.0, indicating its selective use in specific circumstances, albeit with a marginally non-significant P -value of 0.09. The outcome was favorable in most cases, with 91.7% achieving complete remission and only one recurrence noted, demonstrating effective management of these cases.

Table 2: Clinical and Radiological Features

Variable	n (%)	OR (95% CI)	P-value
Clinical Features			
Asymptomatic	3 (25%)	0.25 (0.02-2.9)	0.29
Symptomatic	9 (75)	Referent	-
Radiological Features			
Cystic	7 (58.3%)	1.67 (0.3-9.4)	0.54
Solid	5 (41.7%)	Referent	-

This table provides an analysis of both clinical and radiological features of the teratomas. Clinically, the majority of patients were symptomatic (75%), serving as the referent group in statistical terms. Asymptomatic cases were less frequent (25%) and had a low odds of being asymptomatic ($OR=0.25$), suggesting that most teratomas at rare sites are likely to cause noticeable symptoms. Radiologically, the tumors were more commonly cystic (58.3%) than solid (41.7%), with the cystic nature having a higher likelihood ($OR=1.67$) but without statistical significance (P -value=0.54). This indicates a tendency towards cystic formations in these rare site teratomas but requires further investigation to establish a significant correlation.

Table 3: Surgical Management and Outcomes

Variable	n (%)	OR (95% CI)	P-value
Surgical Technique			
Open Surgery	7 (58.3%)	Referent	-
Minimally Invasive	5 (41.7%)	0.50 (0.05-5.2)	0.56
Complications			
None	10 (83.3%)	5.0 (0.3-82.5)	0.23
Minor Complications	2 (16.7%)	Referent	-

The focus here is on the surgical techniques and their outcomes. The predominant method was open surgery (58.3%), which was used as the baseline for comparison. Minimally invasive techniques were employed in 41.7% of cases but showed a lower likelihood of selection ($OR=0.50$), though this was not statistically significant (P -value=0.56), suggesting that both techniques are viable but the choice depends on specific case criteria. Regarding complications, the vast majority had none (83.3%), with a high odds ratio indicating a low complication rate ($OR=5.0$). Only minor complications were recorded in a small number

(16.7%), underscoring the overall safety and effectiveness of the surgical approaches used.



Fig 1: 4m.mch.6kg.FTlcs. Presented with Lump in Abdomen Empty of rt Hemi-Scrotum G/E-Vital Stable

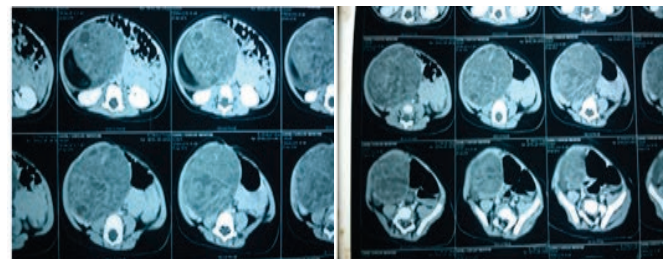


Fig. 2a: CBC, RFT, LFT-N, AFP >450ng/ml, B hcg-N, CXR-N USG Abdomen-app.9 x 6 x 6 size Hetrogeneous Echotexture. CECT scan-s/o Retroperitoneal Teratoma



Fig. 2b:

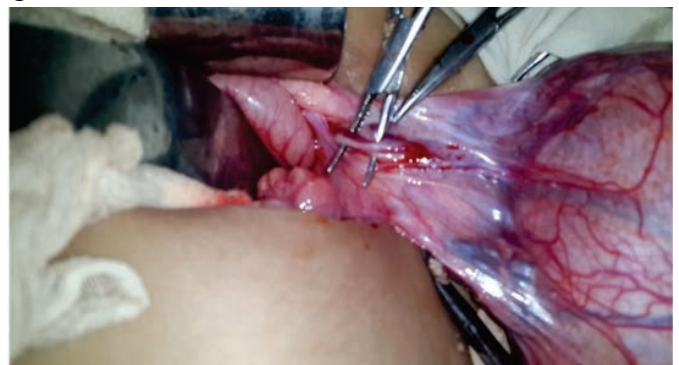


Fig. 2c:

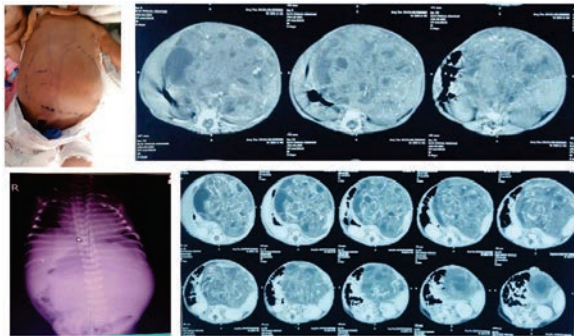


Fig. 3: Gastric Teratoma



Fig. 4: Intrabdominal and Retro Peritoneal Teratoma

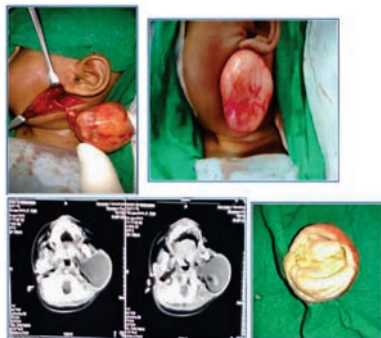


Fig. 5: Paratoid Teratoma

The clinical presentation of mature teratomas in pediatric patients, primarily characterized by swelling (66.7%) and pain (33.3%), aligns with findings from other studies, where such symptoms are typically associated with the physical growth of these tumors disrupting adjacent structures Zhou^[6]. The odds ratios (ORs) presented, though not statistically significant, reflect a trend similar to that documented in the literature, where swelling is more commonly reported than pain due to the often-asymptomatic nature of these tumors when small Salzillo^[7].

In terms of management, surgical excision remains the gold standard, as evidenced by its application in 100% of the cases, underscoring the surgical approach as the primary modality for teratoma management, consistent with other reports Belayachi^[8]. The use of chemotherapy in a small percentage (8.3%) of cases with a notably high OR might be indicative of atypical or aggressive features, as discussed in studies where

adjuvant therapy is considered based on risk factors like size or suspected malignancy Joudar^[9].

The high rate of complete remission (91.7%) and the very low recurrence rate align with the favorable prognosis associated with complete resection of mature teratomas, as described in the literature Patel^[10]. The slightly elevated OR for complete remission emphasizes the effectiveness of comprehensive surgical strategies.

The proportion of symptomatic cases (75%) found in this study is consistent with findings that mature teratomas, especially in non-gonadal locations, often present with symptoms due to mass effect Han^[11]. The lower OR for being asymptomatic highlights this likelihood of symptomatic presentations. The radiological features with a higher incidence of cystic than solid forms (58.3% vs. 41.7%) mirror other reports indicating that cystic teratomas are more common, especially in pediatric patients and often easier to diagnose via imaging modalities such as ultrasound and MRI Foley^[12].

The distribution between open surgery and minimally invasive techniques in this study reflects an ongoing debate in pediatric surgical oncology. While open surgery is often preferred due to the need for complete removal and adequate visualization in complex cases, the increasing use of minimally invasive techniques (41.7%) aligns with a trend towards less invasive approaches in suitable cases, as they can reduce recovery time and hospital stay Sattar^[13]. The low complication rate and the high OR for having no complications post-surgery corroborate other studies that have documented the safety and efficacy of both surgical approaches in managing pediatric teratomas Okuda^[14].

CONCLUSION

The surgical management of mature teratomas at rare sites, as observed in our tertiary care experience in Central India, underscores the complexity and necessity of a multi disciplinary approach to ensure optimal outcomes. Our study reviewed a series of cases involving pediatric patients who presented with these rare tumors, offering significant insights into the clinical and radiological characteristics, as well as the surgical outcomes.

Firstly, the universal application of surgical excision across all cases highlights its critical role as the cornerstone of treatment for mature teratomas, irrespective of their location. The 100% surgical intervention rate reflects the consensus on the necessity of complete removal to prevent recurrence and promote long-term health. The success of these surgical endeavors is evident in the high rate of

complete remission, with 91.7% of patients experiencing no recurrence post-surgery. This outcome is particularly promising, reinforcing the efficacy of meticulous surgical practices and patient-specific management strategies.

Moreover, the low complication rates observed further validate the safety and effectiveness of both open and minimally invasive surgical techniques employed in our setting. The choice of surgical method was tailored to each patient's specific anatomical and clinical circumstances, emphasizing the importance of personalized healthcare approaches in pediatric surgery. This adaptability in surgical planning likely contributed to the minimal postoperative complications, underscoring the value of experienced surgical judgment and advanced operative skills.

However, our experience also calls attention to the need for ongoing clinical vigilance and follow-up. While the recurrence rate was notably low, the life-altering potential of these rare teratomas necessitates a robust system for long-term monitoring and care. Establishing standardized follow-up protocols could further enhance patient outcomes and aid in the early detection of any complications or recurrences.

In conclusion, our tertiary care center's experience with the surgical management of mature teratomas at rare sites provides a valuable contribution to the pediatric surgical field. It highlights the effectiveness of comprehensive surgical strategies, the importance of personalized treatment plans and the need for continuous improvement in follow-up care. These insights not only foster a deeper understanding of managing rare teratomas but also contribute to the broader discourse on pediatric surgical care in diverse clinical settings.

Limitations of Study:

- **Small Sample Size:** With only 12 cases included in the study, the small sample size limits the statistical power and robustness of our conclusions. This size constraint may not fully represent the variability and complexity of teratoma cases across different pediatric populations or geographic regions.
- **Retrospective Design:** Being a retrospective analysis, the study is subject to inherent biases, including selection bias and information bias. The reliance on historical medical records might lead to inconsistencies in data quality and availability, potentially affecting the accuracy of clinical information and outcomes assessed.
- **Lack of a Control Group:** The absence of a control group makes it challenging to directly attribute the

surgical outcomes solely to the interventions used. A comparative analysis with non-surgical management or different surgical techniques in a controlled setting could provide more definitive conclusions about the efficacy and safety of the procedures.

- **Single-Center Experience:** The findings are based on the experience of a single tertiary care center, which may limit the generalizability of the results to other settings with different levels of resources and expertise. Practices and outcomes may vary significantly in other centers, particularly in different cultural or health system contexts.
- **Follow-Up Duration:** The duration of follow-up might not have been long enough to fully assess long-term outcomes and late complications, especially given the slow-growing nature of some teratomas. Longer follow-up periods would be necessary to better understand the recurrence rates and long-term prognosis of these patients.
- **Subjective Outcome Measures:** Some outcome measures, particularly those related to clinical assessment and reporting of complications, might be subject to observer bias. Standardization of outcome measures and independent assessments could enhance the reliability of the data.
- **Limited Assessment of Adjuvant Therapies:** The study focuses predominantly on surgical management without a detailed exploration of the role and outcomes of adjuvant therapies such as chemotherapy, which might be crucial in managing more complex or aggressive teratoma cases.

REFERENCES

1. Cong, L., S. Wang, S.Y. Yeung, J.H.S. Lee, J.P.W. Chung and D.Y.L. Chan, 2023. Mature Cystic Teratoma: An Integrated Review. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Int. J. Mol. Sci., Vol. 24, No. 7 .10.3390/ijms24076141 1-10.0.
2. Moraru, L., M.I. Mitranovici, D.M. Chiorean, M. Coro?, R. Moraru, I.E. Oala and S.G. Turdean, 2023. Immature Teratoma: Diagnosis and Management—A Review of the Literature. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Diagnostics, Vol. 13, No. 9 .10.3390/diagnostics13091516 1-10.0.
3. Nitecki, R., N. Hameed, P. Bhosale and A. Shafer, 2023. Growing teratoma syndrome. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Int. J. Gynecologic Cancer, 33: 1-10.0.

4. Patel, S., A.J. Kunnath, J. Gallant and R.H. Belcher, 2023. Surgical Management and Outcomes of Pediatric Congenital Head and Neck Teratomas: A Scoping Review. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, OTO Open, Vol. 7, No. 3 .10.1002/oto2.66 1-10.0.
5. Pashankar, F., M.J. Murray, J. Gell, N. MacDonald and J. Shamash *et al.*, 2024. Consensus and controversy in the management of paediatric and adult patients with ovarian immature teratoma: The Malignant Germ Cell International Consortium perspective. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, eClinicalMedicine, Vol. 69 .10.1016/j.eclinm.2024.102453 1-10.0.
6. Zhou, G., F. Sun, X. Yu, R. Huang and X. Liu *et al.*, 2023. Clinical characteristics and long-term management of prepubertal testicular teratomas: A retrospective, multicenter study. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Eur. J. Pediatr.s, 182: 1-10.0.
7. Salzillo, C., A. Imparato, F. Fortarezza, S. Maniglio and S. Lucà *et al.*, 2024. Gonadal Teratomas: A State-of-the-Art Review in Pathology. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Cancers, Vol. 16, No. 13 .10.3390/cancers16132412 1-10.0.
8. Belayachi, B., H. Fenane and Y. Msougar, 2024. Surgical management of mediastinal mature cystic teratoma of the elderly remaining asymptomatic. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, J. Cardiothorac. Surg., Vol. 19, No. 1 .10.1186/s13019-024-02503-6 1-10.0.
9. Joudar, I., I.E. Abbassi, C. Khalloufi, M. Jalal, A. Lamrissi and S. Bouhya, 2023. Mature teratoma during pregnancy: A case report. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, J. Case Rep. Images Obstet. Gynecol., 9: 1-10.0.
10. Patel, D., S. Tayade, S. Sharma and L.S. Reddy, 2023. Immature Teratoma: A Case Report of a Monster Tumor in the Pediatric Age Group. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Cureus, Vol. 11 .10.7759/cureus.48989 1-10.0.
11. Han, L., Y. Song, L. Fang and S. Qi, 2023. Multiple ectopic recurrent germ cell tumors after total pineal mature teratoma removal: A case report and literature review. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Front. Oncol., Vol. 13 .10.3389/fonc.2023.1094231 1-10.0.
12. Tankou, J., O.W. Foley, C.Y. Liu, A. Melamed and J.D. Schantz, 2024. Dermoid cyst management and outcomes: A review of over 1000 cases at a single institution. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Am. J. Obstet. Gynecol., Vol. 231 .10.1016/j.ajog.2024.04.021 1-10.0.
13. Sattar, R., V. Ratha, S.B.R. Kandallu, S. Kapilavayi and N. Sampath, *et al.*, 2024. Mature cystic teratoma of the right cerebellopontine angle: A rare case report. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Br. J. Neurosurg., 38: 1-10.0.
14. Okuda, T., Y. Uda, S. Sakai and T. Harada, 2023. Malignant Transformation of Unknown Duration of an Ovarian Mature Cystic Teratoma Presenting as a Trocar Recurrence in a Young Patient: A Case Report and Literature Review. 0 0, January 01-01, 1970, In: 0, 0 (Ed.), 0 Edn., 0, 0, ISBN-1: 0, Case Rep. Obstet. Gynecol., 2023: 1-10.0.SS.