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Index

Sr. No	Title	Author	Subject	Page. No.
1.	Assay Of Triphenylmethane Reductase Enzyme And PCR-Based Identification Of TMR Gene In Enterobacter Asbriae Strain XJUHX-4TM	Tina Mukherjee, Mounita Bhandari, Manas Das	Biotechnology	1-2
2.	An Analysis Of Growth Of Credit Card Industry	Dr. A. Vinayagamoorthy, K. Senthikumar	Commerce	3-5
3.	Impact Of Pre-Merger And Post Merger On Financial Performance (With Reference To Private Sector Banks)	Dr. Shital Vekariya	Commerce	6-8
4.	Relativity On Climate And Competencies In Human Resource Development With Reference To Neyveli Lignite Corporation Ltd,	S. Jayakumar. Dr. R. Ramachandran	Commerce	9-11
5.	Human Resource Outsourcing: A Strategy For Gaining Competitive Advantage	Dr. Santosh M. Singh	Commerce	12-13
6.	Relationship Between EVA And ROI And MVA (A Case Study Of Ten Manufacturing Industries In India)	Dr. Shivani Gupta	Commerce	14-15
7.	Modeling The Traits Of An Effective Teacher At Higher Education	Dr. Haridayal Sharma	Commerce	16-17
8.	Mahatma Gandhi National Rural Employment Guarantee Act (Mgnrega): Issues And Challenges	Dr. Mohd. Ashraf Ali, Mushtaq Ahmad	Commerce	18-20
9.	Standardisation And Grading	Viram. J. Vala, Dr. Vijay Kumar Soni	Commerce	21-22
10.	Profitability Of Selected Information Technology Companies In India	Dr. M. Jegadeeshwaran, C. Udaya	Commerce	23-25
11.	Emerging Trends In The Indian Media And Entertainment Industry	Dr Mahalaxmi Krishnan	Commerce	26-27
12.	Inventory Management Strategies And Control Techniques: An Empirical Investigation Of Small Scale Industries	Vipul Chalotra, Neetu Andotra	Commerce	28-30
13.	A Study On Performance Indicators Of Commercial Banks	Dr. G. Ganesan, P. Parthasarathy	Commerce	31-33
14.	Improved Approaches To Coreference Resolution In Machine Learning	Kuldeep Singh Raghuwanshi, Ashwini Kumar Verma	Computer Science	34-37
15.	Security Issues & Controls In Cloud Computing	V. Naga Lakshmi	Computer Science	38-40
16.	Human Development Index Of De-Notified Nomadic Castes In Maharashtra Division: A Study Of Jalna And Aurangabad Districts	Dr. Ashok Pawar	Economics	41-43
17.	Public Private Partnership In Rural & Urban Projects In India	Dr. Ashok S. Pawar, Dr. Shankar B. Ambhore	Economics	44-45
18.	Populace Insight On Development In Public Health Sector Of India Subsequent To Functioning Of National Rural Health Mission	Krishnakant Sharma	Economics	46-49
19.	Problems Of Rural Women Entrepreneurs In India: A Conceptual Overview	C. Jeyasri Usha N Devi, Dr. A. Sankaran	Economics	50-52
20.	Poverty Of Banjara And Vanjari Communities In India	Tidke Atish S., Dr. Pawar Ashok S.	Economics	53-54
21.	India And China: Economic Reforms And WTO	Dr. Surinder Kumar Singla, Dr. Kulwinder Singh	Economics	55- 56
22.	Implementing Life Skill Education Strategies In Teaching – Learning Process	R. Kalaiselvi, Dr. A. Palanisamy, Dr. A R. Saravanakumar	Education	57-59

23.	Utilisation Of Modern Technology By The Teachers In Pupil Processing Organisation	Dr. P.Paul Devanesan, Dr A. Selvan	Education	60-61
24.	Impact Of Vocational Training On Students	K.Sudha Rani, G.Umapathi, Dr. T. Ananda,	Education	62-63
25.	A Study On Emotional Intelligence Of Secondary School Teachers	Dr. Umme Kulsum, Prathima H.P.	Education	64-66
26.	The Efficiency Of Feedback Strategy Of Homework On The Development Of 10th Grade EFL Writing Skill In Al-Karak Educational Directorate	Majid Al- Khataybeh, Areej Al-Shourafa`	noitacudE	67-74
27.	Perspectives Of Stress Management In Education System	M. Meenakshisundaram, G. P. Raja, Dr. A R. Saravanakumar	Education	75-76
28.	Attention Regulation Of Meditators And Non-Meditators Of Class IX	G. Madhavi Kanakadurga, Dr. D. Vasanta Kumari,	Education	77-78
29.	Role Of Psychoeducation In Teaching – Learning Process	Dr. A R. Saravanakumar, Dr. A. Balu, Dr. S. Subbiah	Education	79-80
30.	Microcontroller Driven RGB Led System For Tristimulus Surface Colorimetry	T. N. Ghorude, A. D. Shaligram	Electronics	81-83
31.	Pmgsy And Rural Roads Development In India: Economic, Financial And Maintenance Issues	K.C. Manjunath	Engineering	84-86
32.	Routing Packets On A Chip.	Naren V Tikare	Engineering	87-89
33.	Finding The Nearest Neighbors In Biological Databases	Er. Pankaj Bhambri, Dr. O.P. Gupta, Er. Franky Goyal	Engineering	90-92
34.	Factors Affecting The Sustainability Of The Asphalt Roads: A Case Study Of Irbid Inner Ring Road, Jordan	Eng. Nasr Ahmad Dr. Mihai Iliescu	Engineering	93-94
35.	Physical And Chemical Testing Of Compounded PVC	Sapna Dabade, Dr. Dheeraj Mandloi, Deepak Khare	Engineering	95-96
36.	Impact Of Organic Farming On Yield Of Some Common Crops- A Case Study.	Namrata D. Awandekar	Environmental Science	97
37.	Hydrogeologic Settings Of The North And South Brahmaputra Plains In Upper Assam: A Comparative Study	Dr. Uttam Goswami	Geology	98-100
38.	To Study Staffing Pattern In Rajasthan Public Healthcare Delivery System.	Dr. Ashwin G. Modi, Sushman Sharma	Healthcare	101-105
39.	Work And Health: A Situational Analysis Of Factory Workers	Dr. S. S. Vijayanchali, Dr. E. Arumuga Gandhi	Home Science	106-108
40.	Performance Of Camel Kid Hair: Acrylic Blended Yarn And Knitted Fabric	Suman Pant, Anjali Sharma	Home Science	109-110
41.	Impact Of Holistic Nutrition Education Package On Diabetes Mellitus Control In Middle Aged Women	Dr. Anjali Rajwade	Home Science	111-112
42.	Assessment Of Relationship Between Ida And Personal Hygiene, Nutritional Knowledge And Dietary Practices In Adolescent Girls	Dr. Anjali Rajwade	Home Science	113-114
43.	Employee Attrition And Retention In Private Insurance Sector– A HRM Challenge	Dr. J. Senthil Vel Murugan, S.Bala Murugan	Human Resource Management	115-117
44.	A Study On Impact Of Unionism On Industrial Relations In Manufacturing Sector	Jaya Ahuja	Industrial Relations	118-120

45.	Augmentation Of India's Foreign Exchange Reserve: An Analysis	Dr.S P.Mathiraj, Ar.Annadurai	International Business	121-123
46.	Films – A Techno Literary Art Form	Dr. Dipti Mehta	Literature	124-125
47.	Indirect Models Of Reading To Develop Descriptive Writing	Dr. K. Madhavi	Literature	126-128
48.	Ramkrishna Mishra Ke Upanaso Me Rajnetaik Chetavni	Dr. Sanjay Rathod, Dilip Jhadav	Literature	129
49.	Hindi Kavita Me Nari Jivan Ka Badla Swarup	Dr. Sanjay Rathod	Literature	130
50.	Impact Of IPL Sponsorship On Consumer Buying Behavior With Reference To Nagpur City	Chandrima Das	Management	131-135
51.	Crowd Sourcing –A New Management Mantra	Devi Premnath, Dr. C. Nateson	Management	136-137
52.	Small Scale Industries In India: An Evaluation Of Productivity In The Post-Liberalized Scenario	Dr. Gaurav Lodha,	Management	138-139
53.	Comparative Analysis Of Milk Products With Respect To Its Competitors With Special Reference To Karnataka Milk Federation (KMF) – At Dharwada City, Karnataka, India	Dr. N. Ramanjaneyalu	Management	140-143
54.	A Study On Work Stress In Women Employees In Coimbatore District	R. Maheswari, N. Brindha	Management	144-145
55.	Accounting For Carbon Credits	Dr. Gaurav Lodha	Management	146-148
56.	A Literature Review On The Relationship Between Training (As A Core Responsibility Of HRM) And Firm Performance.	Priya Sharma, Dr. S. L. Gupta	Management	149-152
57.	A Study On Agricultural Marketing Practices And Constraints With Special Reference To Paddy / Rice.	CM Maran, Dr Raja Pranmalai	Management	153-156
58.	Performance Of Share Price Of Indian Public Sector Banks And Private Sector Banks - Comparative Study	V. Prabakaran, D. Lakshmi Prabha	Management	157-158
59.	Intuitionistic Fuzzy Primary And Semiprimary Ideal	Dr. M.Palanivelrajan, S.Nandakumar	Mathematics	159-160
60.	Significance Of Umbilical Artery Velocimetry In Perinatal Outcome Of Fetuses With Intrauterine Growth Retardation.	Dr G S Shekhawat	Medical Science	161-163
61.	Large Adult Sacrococcygeal Teratoma: A Case Report And Review Of Literature.	Dr.Yavalkar Pa, Dr. Naik Am.	Medical Science	164-165
62.	Epidural Steroid In Low Back Ache	Dr. B. L. Khajotia, Dr. Neelam Meena	Medical Science	166-167
63.	A Comparative Study Of Second Trimester MTP With Use Of Vaginal Misoprostol And Extra Amniotic Instillation Of Ethacridine Lactate.	Dr. Ketaki Junnare, Dr. Sameer Darawade, Dr. Priyamvada Shah, Dr. Swati Mali.	Medical Science	168-169
64.	A Novel Surgical Approach For Treatment Of Sui –TVT Obturator Tape	Dr. Ketaki Junnare, Dr. Durga Karne, Dr Neelesh Risbud.	Medical Science	170-171
65.	Advantage Of Fallopian Tube Sperm Perfusion Over Intra-Uterine Insemination When Used In Combination With Ovarian Stimulation For The Treatment Of Unexplained Infertility.	Dr G S Shekhawat, Dr Pushpalata Naphade	Medical Science	172-175

66.	"Bilateral Sertoli-Leydig Cell Tumor In Postmenopausal Female" A Case Report	Dr. Priyamvada Shah, Dr. Ketakijunnare, Dr. DurgaKarne	Medical Science	176-178
67.	Pretreatment With Ephedrine For Prevention Of Pain Associated With Propofol Injection.	Dr. Kavita U Adate, Dr. Jyoti A. Solanki	Medical Science	179-181
68.	Does The Structured Teaching Programme Influence The Knowledge About Physical Wellbeing Of School Children? A Quasi Experimental Study.	Dr. S. Valliammal, Dr. Ramachandra, Raja Sudhakar	Nursing	182-184
69.	An Approach For Information Retrieval For Bookstores Using Formal Ontology	Sumit Jain, C.S.Bhatia	Ontology	185-187
70.	Analgesic Activity Of Anacardium Occidentale	A. Devadoss, C. Aparna, K. Parimala, D. Sukumar	Organic Chemistry	188-190
71.	Behaviourism : Science Or Metaphysics	Dr. Jatinder Kumar Sharma	Philosophy	191-193
72.	Multi-Dimensional Perspectives Of Obesity And Its Management	S. Dhanaraj, Dr. A. Palanisamy	Physical Education	194-196
73.	Refractive Index, Density, Excess Molar Volume, Excess Molar Refraction For Liquid Mixtures (Ethyl Ethanoate + Benzene Derivatives) At Different Temperatures	Sheeraz Akbar, Mahendra Kumar	Physics	197-199
74.	Refractive Indices, Densities And Excess Properties For Liquid Mixtures (Cetane + Alkanols) At Different Temperatures	Sheeraz Akbar, Mahendra Kumar	Physics	200-202
75.	Capacity Building For Effective Local Governance: Indian Perspectives	Dr. Pralhad Chengte	Political Science	203-205
76.	Psychological Well-Being: A Study Of Non-Institutionalized Aged	Dr. Pankaj S. Suvera	Psychology	206-208
77.	Women Empowerment Through N R E G S (With Reference To State Of West Bengal)	Dilip Kumar Karak	Social Sciences	209-211
78.	Effect Of Selected Yogic, Aerobic And Laughter Exercises On Blood Pressure Of High School Boys	Dr.Manjappa.P, Dr.Shivarama Reddy. M	Sports	212-216
79.	Association Study Between Lead And Copper Accumulation At Different Physiological Systems Of Goat By Application Of Canonical Correlation And Canonical Correspondence Analyses	Partha Karmakar, Debasis Mazumdar, Seema Sarkar (Mondal), Sougata Karmakar	Statistics	217-219
80.	Development Of Silver -Silica Nanocomposite For Novel Humidity Sensing Application	Surender Duhan	Technology	220-221



Large Adult Sacrococcygeal Teratoma: A Case Report And Review Of Literature.

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ABSTRACT

Sacrococcygeal teratomas (SCT) are neoplasm containing tissue derived from more than one primitive germ layer and its cause remains unknown. They (SCT) is one of the most common tumours in infants, but are rare in adults. The incidence in neonates is 1 in 40000 with a female to male ratio of 10:1¹. It is rare in adults with less than a hundred cases being documented in the literature². Most of the sacrococcygeal tumours are cystic and benign with 12% being malignant. Treatment consists of early complete surgical resection, as it has the potential for malignant transformation which increase with age³. We present a case of a large adult sacrococcygeal teratoma in an female and review of its literature. The clinical features, radiological findings and outcome are described.

Keywords : Sacrococcygeal teratoma, presacral teratoma, germ cell tumours



45 years old female presented with a large mass in the right gluteal region. She

had difficulty in sitting, no pain, no bowel or bladder complains. She had noticed the

mass which had slowly grown over the last twenty years to attain the present size. She had not consulted a doctor as it was painless, but due to its large size she had difficulty in sitting. The patient has had a normal obstetric history. On admission we did her routine investigations which were within normal limits. On examination a mass in the right gluteal region measuring about 12 x 6 cms, oval in shape, skin was stretched with a few dilated veins, it was non-tender with firm consistency.

On CT scan a large mass was seen anterior to the sacrum and coccyx compressing the urinary bladder and sigmoid colon and anteriorly displacing the rectum. It was extending inferiorly into the right ischioanal fossa. It was a complex mass with an intact sacrum and no invasion of surrounding structures. The patient was operated under general anesthesia with endotracheal intubation in a jack knife position, a vertical incision was made and a large mass present in the gluteal region was excised. The mass was seen extending up between the sacrum and rectum, this retro rectal mass could be excised by creating a plane between the mass and rectum. Coccygectomy was done to prevent recurrence. Haemostasis was secured and two drains were inserted. Histopathology examination confirmed the diagnosis of benign mature teratoma, with the presence of tissue derived from all three germ layers. There was no evidence of immature cell types or any malignant change. Post-operative recovery was uneventful. The patient was called for follow up every two months in the surgical OPD for the first year. At the end of one year no recurrence was seen on CT scan, she has been advised regular follow up once in six months for another two years.

Discussion

A teratoma is a tumour composed of multiple tissues or cell

types foreign to the site of origin. Sacrococcygeal teratomas (SCT) are neoplasm containing tissue derived from more than one primitive germ layer³ and its cause remains unknown^{4,5}. It is the most common tumour in newborns and the incidence is 1 in 40000 with a female to male ratio of 10:11. It is rare in adults with less than a hundred cases being documented in the literature². The incidence of malignancy increases with age³. Sacrococcygeal area is one of the most common sites for teratoma in the newborn⁶, but in adults the tumors at this site are very rare and seen in the gonads (ovaries and testes), the anterior mediastinum, the retroperitoneal space, the presacral and coccygeal areas, pineal and other intracranial sites, the neck, and abdominal viscera⁷. The tumours may be cystic, solid or mixed^{8,9}. They have been classified morphologically according to their relative extent outside and inside the body by the Altman classification as:-

Type I entirely outside, sometimes attached to the body only by a narrow stalk

Type II mostly outside

Type III mostly inside

Type IV entirely inside; this is also known as a presacral teratoma or retro rectal teratoma.

Little is known about the embryogenesis of SCT, but it has been hypothesized that the anomaly is derived from a primitive knot, also referred to as Henson's node¹⁰. SCTs are thought to originate from multipotential cells in Henson's node, which migrates caudally to rest in the coccyx. Various other theories exist to explain the origin of SCT. These include nonsexual reproduction of germ cells within the gonads or in extragonadal sites; 'wandering' germ cells of non-parthenogenetic origin left behind during the migration of embryonic germ cells from yolk sac to gonad; or origin in other totipotential embryonic cells¹¹.

Sacrococcygeal teratomas (SCT) are the most frequent solid congenital tumours in the foetus and the newborn. It is rare in adults with only a few published cases in literature¹². The sacrococcygeal area is the most frequent site of teratoma in infancy. This tumor rarely presents in adulthood, and is

A female, rather than male, predominance exists^{2,12} with a ratio of about 10:11^{2,13}. The clinical presentation varies on the location and extent and symptoms are related to mass effect such as back pain, bowel & bladder dysfunction or they may have neurological symptoms like lower extremity numbness^{10,11}.

Biochemical markers like AFP, carcinoembryonic antigen, and HCG may be elevated in patients with malignant lesions and also help in detection of recurrence after surgery¹⁴.

Large mass may cause displacement of the uterus and rectum which can be felt on digital examination. Patients may also present with abscess or perirectal fistulas¹⁵. Unlike teratomas in infants, which are externally visible in 90% of cases, sacrococcygeal teratomas in adults are confined mostly to the intrapelvic space¹⁶. There is a tendency in the neonates toward malignant transformation of SCTs with increasing age¹⁷. However in adult patients benign tumors predominate¹⁸. Adult patients with SCT may be asymptomatic with the diagnosis being an incidental finding in a digital rectal examination or as a retro-rectal mass on CT. Differential diagnoses are anterior meningocele, rectal duplication cysts or anal gland cysts. Complete surgical excision is the treatment of choice. Excision of the coccyx may be necessary because the bone may contain a nidus of pluripotent cells with a risk of recurrence¹⁹. For benign disease, no further adjuvant therapy may be necessary. Traditional treatment of sacrococcygeal teratomas favors always removing the coccyx with the tumor, since failure to do so results in an approximately 30% to 40% recurrence rate. Many case reports fail to recognize the important distinction between isolated presacral teratomas (defined here as those with no apparent tissue attachment to the coccyx) and true sacrococcygeal teratomas which have attachments to the coccyx.

Symptoms specific to the sacrococcygeal and presacral tumor locations are those of mass effects displacing contiguous structures, rectal pain, backache, paresthesias,

numbness in the so-called saddle area and legs, and loss of bladder and rectal control. Failure to differentiate between the two entities could lead to unnecessary coccygectomies. Complications seen with coccygectomy are delayed wound healing

with increased wound infection rate, increased recovery time, and rarely, bowel

herniation^{20,21}. Access to the tumour could be by transabdominal approach or the transperineal route using the jack-knife position or a combined approach. Complete excision has extremely good prognosis, and is necessary to prevent local recurrence and potential malignant transformation. The sacrococcygeal approach or combined abdominal-sacral approach is recommended²².

Conclusion

Although rare in adults, sacrococcygeal teratoma should be considered in the differential diagnosis of patients with a pelvic mass presenting with obstructive symptoms. Long-term survival is possible with adequate resection of the tumor. The presence of leptomeningeal involvement and malignant transformation are associated with a less favorable outcome. Surgical removal is generally indicated at the time of detection, as these lesions carry a significant malignant potential. Differentiation between presacral teratoma and sacrococcygeal teratoma is important to prevent unnecessary coccygectomies.

Large sacrococcygeal teratoma in adult



Intra op

Post-op



Ct scan



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