International Journal of Science and Research (IJSR)
ISSN: 2319-7064
SJIF (2022): 7.942

Unusual Presentation of Congenital Dermal Sinus on Lateral Buttock

Dr. Sagar B Joshi[1], Niharika N Singh[2]

[1]MBBS, GMERS Medical College, Gotri, Vadodara, Gujarat, India 390021, +91 8154881736
Corresponding Author Email: *drsagarjoshi16[at]gmail.com*
+91 8154881736

[2]MBBS, GMERS Medical College, Gotri, Vadodara, Gujarat, India 390021, +91 8140800085
Email: *niharika16ns[at]gmail.com*

Abstract: ***Congenital Dermal Sinus is rare disease usually present in the midline of the body and may communicate with spinal canal. We present one-year-old boy with dimple inthe right lateral buttock noted since birth. MRI showed tract extending from dimple, traversing the subcutaneous tissue and connecting to the abscess cavity in right pelvic cavity and not connected to the spinal canal. MRI also showed no abnormality in spinal canal. Histopathological examination had shown sinus tract lined by squamous epithelium suggesting epidermoid cyst. Staph aureus (MSSA) and Klebsiella pneumoniae species are identified in culture.***

Keywords: Congenital Dermal Sinus, Lateral Buttock

1. Introduction

Congenital Dermal Sinus is rare disease which usually present in the midline of the body and may communicate with spinal canal [1] [2] [3]. In an extremely rare case, it may present in the lateral side and may end blindly without communicating with spinal canal [4]. Usually, located in the posterior midline from occiput to lumbosacral region, most cases found between lumbosacral region [5]. Nature of CDS is a retro-rectal developing dermoid cyst or epidermoid cyst and total excision of the abnormal tract is the only treatment [5]. We are describing a case of lateral Congenital Dermal Sinus in the gluteal region and illustrating neuro-imaging findings of this extremely rare entity.

2. Case Study

A one-year-old boy presented with low grade fever and purulent discharge from a dimple in the right lateral buttock (FIGURE 1). Physical examination revealed pus discharge from above mentioned site, fluid collection palpated. Laboratory finding showed elevated WBC and CRP and Staphaureus (MSSA) and Klebsiella pneumoniae species are cultured from the pus. The paediatric surgeon suspected infected Congenital Dermal Sinusand prescribed imaging studies.Magnetic resonance imaging(MRI) revealed a tract that extending from the dimple in the right buttock,extends cranially and medially, then abuts superior margin of right gluteus maximus muscle, curves down and enters greater sciatic foramen, then enters right pelvic cavity and end blindly in a well-defined peripherally loculated/abscess in right pelvic cavity but no abnormality in the spinal canal (FIGURE 2). Computed tomography(CT) showed the hypodense lesion in the retroperitoneal plane of right side of pelvis with surrounding fat density also extending to the right gluteal region(FIGURE 3). Patient operated for the same and histopathological studies had done. Patient presented again after couple of weeks with same history, shown recurrent abscess formation. Subsequent CT showed minimal increase in the soft tissue in the subcutaneous plane of the right gluteal region with minimal inflammation of surrounding fat however there is minimal reduction in the dimension of the hypodense lesion/collection in the retroperitoneal plane of the right side of the pelvis. Histopathological examination showed that the sinus was lined by Stratified Squamous Epithelium. Additionally imaging findings also revealed right kidney at ectopic in location seen in right side of pelvis and small sized left kidney and 4mm gall bladder calculus which may associate with congenital anomaly.

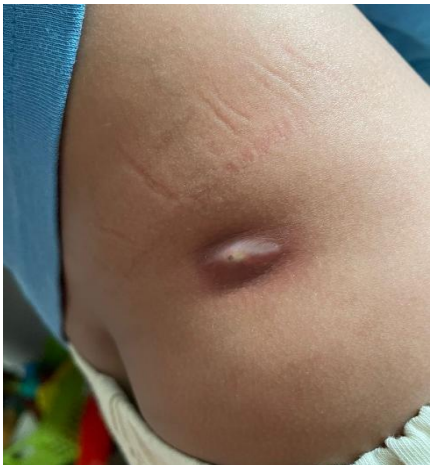

Figure 1: Dimple on the skin of right lateral buttock

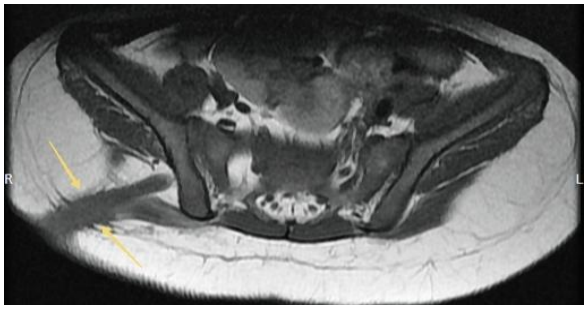

Figure 2: MRI showing a fistula extending from right lateral buttock to right pelvic cavity.

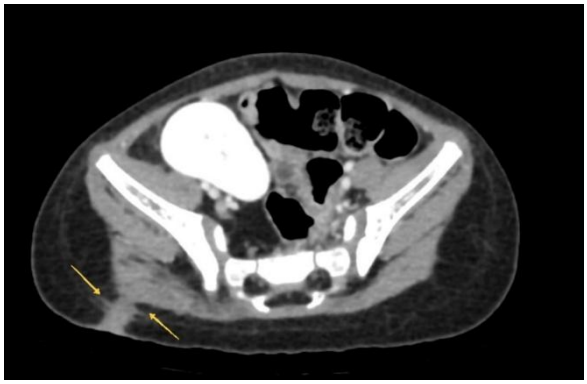

Figure 3: CT showing tractfrom skin extendinginto right pelvic cavity

3. Discussion

Congenital Dermal Sinus usually situated on the midline and communicate with spinal canaland lined by stratified squamous epithelium. Lateral Congenital Dermal Sinus in the buttock is an extremely rare lesion with very few numbers of cases reported. Congenital Dermal Sinusis the failure of the neuroectoderm to separate from the surface ectoderm during the process of neurulation [5].Embryologically, because of failure of separation of neuro ectoderm with surface ectoderm,surface ectoderm wanders into the neural tube and forms a fistula; it is the period of neural tube formation at around 3–4-week [5] [6]. The clinical features of the few cases including our case are shown in Table 1.1. The aetiology of such rare cases has not yet clearly understood but several hypotheses have been proposed. i.e., Hypothesis-1 - Lateral dermal sinus occurs during the process of dermoid cyst formation. Dermoid cyst usually appears along the lines of embryonic fusion during fetal development, and it considered that some pieces of ectoderm pinched off as the suture lines close and become sequestrated from the cutaneous tissue. During this if surface ectodermal pieces do not get pinched off completely and form tract before they separate from the surface skin, a dermal sinus is formed. If this maldevelopment occur on lateral side of the body, a lateral dermal sinus occurs like an epidermoid cyst (Epidermoid cyst: not situated in midline and does not contain skin adnexa) [7]. Hypothesis-2 – Zipping Error; according to these two mechanisms may be responsible. Out of which one is misalignment of neural fold during fusion between the ascending and descending closure region of Neural Tube and second one is mass lesion such as lipomatous tissue occupy the closure tract of the neural fold and leave some excess neural folds around them laterally [8]. A case of infected lateral dermal sinus can be mis-diagnosed as benign skin lesion such as atheroma or skin acne. Congenital Dermal Sinus candiagnose with imaging studies i.e., USG, CT, MRI, Contrast study.The aim of treatment is to complete resect the abnormal tract without injuring neural structure as it may communicate with spinal canal. Prior to surgical management is it very essential to detect the precise distal end of sinus located from the orifice for which above mentioned imaging study may help.

Table 1.1: Clinical features of atypical cases

Author (Year)	Age	Sex	Side	Distal end of the sinus
Rigg et al. (1975) [9]	22 years	F	L	Duplication cyst and peritoneal cavity, extraspinal canal
Carrillo et al. (1985) [10]	22 months	M	L	Subarachnoid, intraspinal canal
Ikwueke et al. (2008) [6]	3 years	F	R	Gluteal fascia, extraspinal canal
Qi et al. (2010) [3]	3 years	F	L	Tip of the coccyx, extraspinal canal
Yamaguchi et al. (2011) [5]	6 months	F	R	Paravertebral fascia, extraspinal canal
Nishimon et al. (2014) [11]	14 years	F	R	Periosteum of right sacral ala, extraspinal canal
Hosokawa et al. (2020) [12]	1 years	F	R	Anterior sacrum, extraspinal canal
Tamotsu Kobayashi et al. (2021) [4]	1 years	M	R	Periosteum of right sacral ala, extraspinal canal
Present case	2 years	M	R	Right pelvic cavity

4. Conclusions

The present case was extremely rare Congenital Dermal Sinus opening in the right lateral buttock. Congenital Dermal Sinuscan be present other than midline lesion and can be diagnosed with imaging studies i.e., USG, CT, MRI, Contrast medium study. For cure complete resection of the abnormal tractis needed with the precaution of not to injure neural structure nearby, hence precise pathway should be detected prior to definitive surgical intervention.

References

[1] Shah, R. K., Chaljub, G., &Swischuk, L. E. (2003). Lower cervical dermal sinus tract and associated intraspinal abscess causing meningitis in a child. Emergency Radiology, 10, 160-162.

[2] Mack, W. J., &Ghatan, S. (2007). Congenital pterional dermal sinus in an 18-month-old child: case report. Neurosurgery, 61(3), E661.

[3] Qi, S., Zheng, J., Lü, Z., Li, K., Chen, L., & Xiao, X. (2010). Lateral buttock and postanal congenital dermal sinus tract. Journal of Pediatric Surgery, 45(5), e23-e25.

[4] Kobayashi, T., Kanamori, Y., Furugane, R., Mori, T., Kano, M., Takahashi, M., &Fujino, A. (2021). Congenital dermal sinus in the lateral buttock. Journal of Pediatric Surgery Case Reports, 75, 102087.

[5] Yamaguchi, S., Takeda, M., Kihara, H., Eguchi, K., Mitsuhara, T., Matsushige, T., &Kurisu, K. (2011). Lateral Buttock Congenital Dermal Sinus Tract—Case Report—. Neurologia medico-chirurgica, 51(6), 460-462.

[6] Ikwueke, I., Bandara, S., Fishman, S. J., & Vargas, S. O. (2008). Congenital dermal sinus tract in the lateral buttock: unusual presentation of a typically midline lesion. Journal of pediatric surgery, 43(6), 1200-1202.

[7] Kubo, T., Takagi, T., Yamaguchi, Y., Yano, K., & Hosokawa, K. (2008). Congenital peristernal dermal sinuses: a new entity?. British Journal of Dermatology, 159(3), 763-765.

[8] Lee, C. S., Phi, J. H., KiM, S. K., Cho, B. K., & Wang, K. C. (2009). Spinal congenital dermal sinus with dual ostia. Journal of Neurosurgery: Pediatrics, 3(5), 407-411.
[9] RIGG, B. M., & MARTYN, J. W. (1975). An unusual congenital dermal sinus of the buttock. Plastic and Reconstructive Surgery, 56(5), 581-583.
[10] Carrillo, R., Carreira, L. M., Prada, J. J., & Rosas, C. (1985). Lateral congenital spinal dermal sinus: a new clinical entity. Child's Nervous System, 1, 238-240.
[11] Nishimon, M., Shimizu, Y., Ueno, M., & Iwanami, A. (2014). Late-onset congenital lateral dermal sinus tract. Case Reports, 2014, bcr2014206530.
[12] Hosokawa, T., Tanami, Y., Sato, Y., & Oguma, E. (2020). Image findings of dermal sinus in the lateral buttocks. Radiology Case Reports, 15(5), 586-591.