



Congenital Absence of Coccyx: A Case Report

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ABSTRACT: Caudal Regression Syndrome is a rare congenital abnormality of lower lumbar spine, sacrum and coccyx. It also includes anomalies of genitourinary system, neurological impairment in lower limb and anorectal anomalies. Here we present a rare case of rudimentary coccyx along with completely open sacral canal from S1 to S5 segments in a child of seven years born to a non-diabetic mother.

KEY WORDS: Caudal Regression Syndrome, Absent Coccyx.

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I. INTRODUCTION

Lower lumbar abnormality like absence or agenesis of sacrum and coccyx have been grouped as Caudal Regression Syndrome. It is a rare congenital abnormality seen usually in a diabetic mother. Such cases are also having genitourinary abnormality and neurogenic bladder. Here we are presenting a girl child of seven years with rudimentary coccyx with completely open sacral canal without any other abnormality of genitourinary system, born to non diabetic mother.

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II. CASE HISTORY:

A girl child aged seven years attended Ortho OPD, Vivekananda Polyclinic and Institute of Medical Sciences, Lucknow with chief complain of severe pain in buttock and lower back while sitting on wooden chair of her class room for the last few months. She is the only child in the family of low socio-economic status. Her mother got this child after 16 years of marriage while she was of 34 years. Her mother is normotensive and nondiabetic. Mother of girl child was having normal antenatal, natal and post natal period. Her milestones were normal, She is of normal built and weight as per her age. Her ADL including ambulation arc normal. Her buttocks arc of normal contour with normal curvature of spine. The neurological examination of her all the four extremities were within normal limit. She can freely squat, sit cross legged, jump and run without any discomfort. She was having diffuse discomfort in lower sacral region, without any swelling or sinus. Her organic reflexes are normal. She has no pain in standing or walking.

Investigation: Her routine blood tests were normal. Ultrasound of lower abdomen revealed no abnormality in genitourinary system ie. both kidney, ureter and urinary bladder.

Digital Xrays of sacro-coccygeal spine-antero-posterior and lateral view (Fig No.1) showed absence of coccyx and small sacrum. CT Scan of sacro-coccygeal spine (Fig No-2) showed the following features-

- All five piece of sacrum are visualised.
- There is completely open sacral canal starting from S1 segment to S5 segment .
- The coccyx is rudimentary and two small rudimentary pieces of coccyx are seen (usually there are 3-4 pieces of coccyx)
- No soft tissue lesion seen posterior to the open sacral canal.

Impression- CT Scan of sacrum and coccyx shows

- 1) Type I posteriorly open sacral canal (open from S1 to S5 level).
- 2) Extremely rudimentary coccyx.

Type I open sacral canal usually presents with pain over the lower sacrum.

III. DISCUSSION

Caudal Regression Syndrome has been documented by various researchers (1 to 5). Joshi and Yadav (2005) reported CRS as a uncommon congenital malformation in the general population occurring in approximately 1 of 25000 live birth,

Bhasin, Jalali and Mechdivatta (1968) reported congenital absence of coccyx as partial or complete. They have quoted Foix and Hillmand (1924) who have classified sacrum and coccyx absence into four types as complete sacrococcygeal agenesis (Type I), subtotal Sacro-coccygeal agenesis (Type II), absence of external half of sacrum (Type III) and total or partial agenesis of coccyx (Type IV).

The present case is of fourth type as described above. Where as there are usually no symptoms and diagnosis is usually made when routine Xrays are undertaken. In the present case child presented as a case of coccydenia ie. pain around coccyx while sitting on hard surface. Pain was relieved in lying and standing. She was advised sitz bath, avoid constipation and use foam cushion for sitting on wooden chair. Her discomfort was slowly reduced. Her ultrasonography of lower abdomen revealed no abnormality in genito-urinary system. Some of CRS cases reported these anomalies as well(Joshi &Yadav,2005). Jadav, Gandhi, Soni and Desai (2012) reported history of delay in toilet training in a case of CRS but such delay in toilet training was not present in the present case. We have not found such case in literature having completely open sacral canal with rudimentary coccyx, without any genitourinary and other skeletal anomalies born to nondiabetic mother. However we have explained the prognosis to her mother.

Summary:

A child having discomfort in sitting on hard surface should be thoroughly evaluated clinically and radiologically to exclude Caudal Regression Syndrome. In present case, radiologically rudimentary coccyx was noted without any other associated abnormality.

Conflict of interest: Nil.



Figure No 1 (Digital Xray of sacro-coccygeal spine Antero-Posterior and lateral views- absence of coccyx)



Figure No 2. CT Scan of sacro-coccygeal spine with 3D showing-

- 1) Type I posteriorly open sacral canal (open from S1 to S5 level).
- 2) Extremely rudimentary coccyx

REFERENCES

- [1]. Col K K Sen, Surg Lt M Patel, "Caudal Regression Syndrome", *Mil.J.Armed forces of india*,63,178-179,2007
- [2]. Mari M. Boulas, "Recognition of CRS", *Advances in Neonatal Care*, Vol 9, No2 PP 66-69, 2009
- [3]. Ali Al Kaissi, K Paus Klaushofer and Franz Grill, CRS and popliteal webbing in connection with maternal DM: A Case Report and literature Review, *Cases journal*:2008 <http://www.casesjournal.com/content/1/1/407>
- [4]. Kei Samba and Kon-ichi Yamamura, "Etiology of Caudal Regression Syndrome", *Human Genet Embryol*, 3:107, May 2013
- [5]. Joshi M, S.Yadav, "Lumbosacral Agenesis", *Ind.J.Radiol Jmag* 15:2:251-254,2005
- [6]. Vipashyana J.Jadav,Jenny Gandhi, Himanshu C.Soni, Saumit Desai, "Caudal Regression Syndrome", *Gujarat Medical Journal*,Vol 67,No2 Aug2012
- [7]. Foix C and Hillemond P, *Rev, Neural*, 31:450,1924. As quoted by RP Bhasin, BK Jalali, KK Mehdimatta.
- [8]. Bhasin RP, Jalali BK, Mehdiratta KK, "Congenital absence of Coccyx", *Indian Paed*,Vol 5, No.1, March 1968

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