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## RIBS IN THE SACROCCOCCYGEAL REGION

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Supernumerary ribs are congenital anomalies that can occur in all areas of the vertebral column. Sacroccocygeal ribs, arising from the most caudal part of the vertebral column, are the rarest type of supernumerary ribs. They were described as early as 1960, with new reports being published every few years. Brief literature overviews were sometimes included with case reports. However, until now, no comprehensive reviews with reported search strategies have been published.

*The purpose of the study* was to present a comprehensive review on the subject. A proposal for a clear definition of these entities is made. Where relevant, comparisons with other types of supernumerary ribs are made. An overview of supernumerary ribs in other mammals is presented.

*Materials and methods.* A search of the Medline and Web of Science Core Collection databases was performed with several relevant keywords. The Google search engine was utilized to identify relevant literature. The references of the publications identified with the former methods were also searched. The publications were sorted according to whether they fit the definition of sacroccocygeal ribs put forward above or were called sacroccocygeal ribs but did not fit the definition. To avoid missing any relevant literature, all literature mentioning sacroccocygeal ribs regardless of the definition used were included. Additionally, literature about pelvic digits that would fit the definition was also included. Papers were excluded if they could not be sourced or did not mention sacroccocygeal ribs (any definition) or pelvic digits fitting the definition.

*Results and discussion.* Sacroccocygeal ribs seems to occur equally often in men and women, since both accounted for half of the twelve cases identified that fit the definition. On average, they are diagnosed at 29.91 years of age, but this varied widely (from 3 to 64). This variation is to be expected, since the entities are usually discovered by accident and are present since birth. Although they are very rare, no further information about risk factors was identified. Most of the patients in the case reports did not suffer from symptoms relating to their sacroccocygeal ribs. However, some authors mention that the ribs might pose obstetrical problems during childbirth. This can be expected to be dependent on the exact size and anatomical location of the rib in question and may vary between patients.

Due to the lack of symptoms that are associated with sacroccocygeal ribs, an underdetection could however be present. Therefore, further research is needed to more specifically assess the prevalence. This limited body of evidence makes it difficult to make sound conclusions about the epidemiology of these entities.

*Conclusion.* Based on the current body of evidence, it is difficult to make sound conclusions about the condition. It is however very clear that the anomaly is very rare and that complications are relatively rare.

**Keywords:** sacroccocygeal, sacral, coccygeal, supernumerary, ribs.

**Introduction.** Supernumerary ribs are congenital anomalies that can occur in all areas of the vertebral column [1-3]. The most common type are cervical ribs that have been estimated to occur in 0.05-3 % of the population [4, 5]. These entities have been relatively well studied. Their clinical implications, such as thoracic outlet syndrome, associated symptoms and possible treatment strategies have all been described [5, 6]. Despite being rarer, there have also been numerous case reports of supernumerary intrathoracic ribs [7-9]. Their aspects were recently described in a literature review by Muise and colleagues [9]. Like intrathoracic ribs, lumbar ribs are also very uncommon. A review article published in 2015 summarized various aspects [10]. They are often asymptomatic and discovered by accident during investigations for other reasons [8].

Sacroccocygeal ribs, arising from the most caudal part of the vertebral column, are the rarest type of supernumerary ribs. They were described as early as 1960 [11], with new reports being published every few years. Brief literature overviews were sometimes included with case reports. However, until now, no comprehensive reviews with reported search strategies have been published.

Several terms are used for sacroccocygeal ribs and related entities. These include, amongst other: pelvic digits, pelvic ribs, pelvic finger, eleventh finger, sacral ribs, coccygeal ribs and iliac ribs. Some of these terms are used interchangeably, while others lack a clear definition. We propose to use the term pelvic ribs only for entities directly connected to the vertebrae of the sacrum and coccyx. Pelvic ribs con-

necting to the vertebral column at these locations can then be called sacral – and coccygeal ribs respectively. Additionally, they should have a congenital origin. This way, these entities can be morphologically linked to cervical, intrathoracic and lumbar ribs. The term pelvic digit can then be reserved for bony structures that do not directly connect to the vertebral column and may originate by other means than congenital malformations. Examples of such structures would be pelvic digits arising from myositis ossificans after trauma.

Currently, not enough cases are known to develop a more specific classification. However, when enough data is available, a specific classification would be developed. This classification could take into account factors like uni- or bilaterality of the ribs, the vertebra where the ribs originates, grade of development, course of the ribs and potential symptoms. An example of such a classification could be the one developed by Kamano and colleagues for intrathoracic ribs [7].

**The purpose of the study** was to present a comprehensive review on the subject. Additionally, a proposal for a clear definition of these entities is made. Where relevant, comparisons with other types of supernumerary ribs are made. Finally, an overview of supernumerary ribs in other mammals is presented.

**Search strategy and inclusion.** A search of the Medline and Web of Science Core Collection databases was performed with several relevant keywords such as 'sacral ribs', 'coccygeal ribs' and 'sacroccygeal ribs'. Additionally, the Google search engine was utilized to identify relevant literature. The references of the publications identified with the former methods were also searched. The publications were sorted according to whether they fit the definition of sacroccygeal ribs put forward above or were called sacroccygeal ribs but did not fit the definition. To avoid missing any relevant literature, all literature mentioning sacroccygeal ribs regardless of the definition used were included. Additionally, literature about pelvic digits that would fit the definition was also included. Papers were excluded if they could not be sourced or did not mention sacroccygeal ribs (any definition) or pelvic digits fitting the definition.

It was clear that the literature about these entities is very scarce. A total of nine case reports fitting the sacroccygeal rib definition were identified (**Table 1**), describing a total of twelve unique cases. These cases accounted for fourteen sacroccygeal ribs, including one bilateral pair and both a unilateral sacral and coccygeal rib in the same individual. Eleven sacral and three coccygeal ribs were described. An additional three case reports mentioning sacral ribs not fitting the definition were identified. No other type of literature, such as larger case series or prospective

studies were reported. Several case reports did however mention and describe previously published cases. The large majority of included papers exclusively mentioned sacral ribs. A further two case reports were identified [12, 13], but could not be sourced.

## Results.

### Demographics of patients and epidemiology

Sacroccygeal ribs seems to occur equally often in men and women, since both accounted for half of the twelve cases identified that fit the definition. This is in contrast with a study that found that cervical ribs occur more often in females than males [4]. For both types of entities, these results are, however, based on a small number of cases identified. Their average age at diagnosis was 29.91 years, but this varied widely (from 3 to 64). This variation is to be expected, since the entities are usually discovered by accident and are present since birth.

Based on the very limited number of case reports, it is clear that sacroccygeal ribs are extremely rare. No prevalence can be calculated based on the limited body of evidence. It does however seem that these entities are rarer than cervical ribs, which have an estimated prevalence of between 0.05-3 % [4, 5] and lumbar ribs with a prevalence of around 1 % [10]. However, caution should be applied when calculating a more precise prevalence of sacroccygeal ribs, since the lack of problems associated with their presence may lead to an underdetection. This could cause an underestimation of their prevalence.

The identified publications did not provide additional information to assess potential epidemiological or risk factors that could be linked to the occurrence of sacroccygeal ribs. Supernumerary ribs in the cervical and lumbar region are however often found in developmental toxicology bioassays [14]. Therefore, there may be toxicological risk factors for developing sacroccygeal ribs. This hypothesis does require a lot more research, which may be unfeasible based on the very rare nature of the entity. The presence of almost all sacroccygeal ribs was confirmed using radiography, with a single case being confirmed with MRI.

### Anatomical characteristics of sacroccygeal ribs

Of the sacral ribs, one originated at S1, one at S2, two at S4 and one at S5. One rib originated at S3 or S4. For most of the sacral ribs, the length was not mentioned. Where it was reported, the length ranged from 10 to 15 cm. For only one rib a width was mentioned, which was 1 cm. Several ribs were mentioned to project towards the ischial bone. Two ribs were mentioned to extend towards the gluteal muscles. One rib was described to run caudally in an anterior direction, while another one ran laterally and caudally. 9 out of 10 cases described unilateral ribs, while one

Table 1. – Overview of case reports and extracted data

Reference	Age	Gender	Laterality	Diagnostic technique	Symptoms	Locality	Anatomical characteristics	Treatment
<b><u>Ribs according to definition</u></b>								
Bohutova et al., 1980	40	Female	Unilateral - right	Radiography	Dysuria (Non-causative)	Sacral	Origin: S1 15 cm long 1 cm broad	Surgical removal
Bohutova et al., 1980	64	Male	Bilateral	Urogram	None	Sacral	Origin: S5 Caudal projection to ischial bones	No surgical removal
Carbo et al., 2015	36	Male	Unilateral - left	Radiography CT	Renal colic symptomatology (likely linked to kidney stones)	Sacral	Origin: S4 10 cm long Downward pathway - anterior route	No information
de Miguel et al., 2007	12	Female	Unilateral - left	Radiography	Hip pain (walking, sitting)	Sacral	Extended into gluteal muscles	Surgical removal
de Miguel et al., 2007	6	Female	Unilateral - left	Radiography	None	Sacral	No information	No surgical removal
Van Derslice et al., 1992	31	Male	Unilateral - left	Radiography	Low-back pain (due to fall)	Sacral	Origin: S2 12 cm long From S2 to greater trochanter	No information
Dunaway et al., 1983	20	Male	Unilateral - right (sacral) Unilateral - right (coccygeal)	Radiography Radiography	None	Sacral Coccygeal	<u>Sacral rib</u> Origin: S3/4 Extends laterally and caudally <u>Coccygeal rib</u> Origin: Co2/3 Extends laterally and caudally	No surgical removal No surgical removal
Miyakoshi et al., 2013	17	Female	Unilateral - left	Radiography CT	Low back pain Discomfort in bilateral gluteal region	Sacral	Origin: S4 55 x 25 x 15 mm Extends obliquely toward the left ischial spine	Conservative treatment No surgical removal
Muddassir et al., 2008	45	Male	Unilateral - right	Radiography	Right flank pain	Sacral	«finger-like bony protuberance»	No surgical removal
Pais et al., 1975	55	Female	Unilateral - right	Radiography	None	Coccygeal	Origin: Terminal coccygeal segment Extends laterally and caudally, termination in the gluteal region. Well differentiated	No surgical removal
Pais et al., 1975	«Middle aged»	Male	Unilateral - right	Radiography	None	Coccygeal	Extends laterally and caudally Small Normally differentiated	No surgical removal

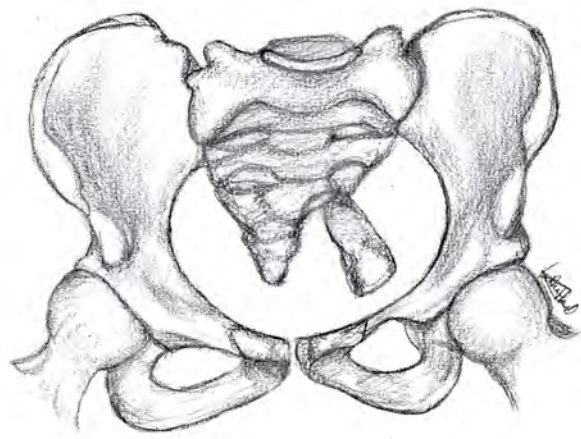
End of table 1

Reference	Age	Gender	Laterality	Diagnostic technique	Symptoms	Locality	Anatomical characteristics	Treatment
Shah et al., 2006	3	Female	Unilateral - left	MRI	Urinary incontinence Bowel incontinence Difficulty walking	Sacral	Origin: S1 Extends behind SI joint into gluteal region Linear Moderately large	No surgical removal
<b>Ribs by name</b>								
Halloran, 1960	49	Male	Bilateral	Radiography	Hip pain (radiating to legs and ankles)	Sacral	Extends bilaterally to greater trochanter	Surgical removal
Heligman et al., 1987	14	Female	Unilateral - right	Radiography	None	Sacral	<u>Rib 1</u> Origin: S1 <u>Rib 2</u> Origin: S4	Partial surgical removal
Mares and Bar-Ziv, 1981	4.5	Female	Unilateral - right	Radiography	None	Sacral	Origin: S4/5 Extends obliquely towards ischial spine	No surgical removal

described bilateral ribs. Of the 9 unilateral ribs, 6 were present on the left side and 3 on the right.

Regarding the coccygeal ribs, one rib was mentioned to originate from the second or third coccygeal segment, while one was stated to originate from the most caudal segments. All coccygeal ribs were stated to extend laterally and caudally. Two of the ribs had a normal differentiation. All coccygeal ribs were unilateral and occurred on the right side.

A representative anterior view (**Figure 1**) and lateral view (**Figure 2**) of a sacral rib was constructed based on images of all studies included.



**Figure 1.** Anterior view of a sacral rib



**Figure 2.** Lateral view of a sacral rib

#### Symptoms and treatment

Most of the patients in the case reports did not suffer from symptoms relating to their sacrococcygeal ribs. However, some authors mention that the ribs might pose obstetrical problems during childbirth [3, 15]. This can be expected to be dependent on the exact size and anatomical location of the rib in question and may vary between patients.



Certain symptoms were mentioned in the publications regarding sacral ribs, such as dysuria, renal colic symptoms, hip pain and low-back pain. These symptoms were however often explicitly mentioned to be caused by other problems or are unlikely to be caused by the presence of sacrococcygeal ribs. No symptoms were reported in patients with coccygeal ribs.

#### Supernumerary ribs in other mammals

Like in humans, supernumerary ribs are very rare in several other mammals. A pair of bilateral cervical ribs was described in a 4-year old female dog in 2017 [16]. Another pair of bilateral cervical ribs was described in a Doberman Pinscher [17]. This animal was female and 11 years old. Unilateral cervical ribs were also described in 2 cats [18]. Additionally, this retrospective study mentioned lumbar ribs in cats. Finally, cervical ribs were described in the extinct woolly rhinoceros [19]. No literature describing supernumerary intrathoracic, lumbar, sacral or coccygeal ribs in other mammals was identified.

#### **Discussion**

Sacrococcygeal ribs seem to be very rare, with only selected case reports being published in the literature. Due to the lack of symptoms that are associated with this entity, an underdetection could however be present. Therefore, further research is needed to more specifically assess the prevalence. This limited body of evidence makes it difficult to make sound conclusions about the epidemiology of these entities. However, based on these case reports, sacrococcygeal ribs seem to occur equally in males and females. They are discovered at a wide range of ages, usually as an incidental finding. The large majority of cas-

es present as unilateral ribs that can originate from any sacral or coccygeal segment. This is in contrast with cervical ribs, which occur most often on C7 [20]. These entities seem to rarely cause symptoms and are usually discovered as an incidental finding. Some case reports however mentioned potential risks for childbirth.

The first step in better understanding these entities in the future is to establish a clear definition. The need to distinguish between sacrococcygeal ribs and pelvic digits has been raised before [3]. A prospective study to identify these anatomical variations is neither feasible nor ethical due to their rarity and the risks associated with the radiological methods by which these entities are usually discovered. However, a retrospective study of images taken of the pelvic region during routine practice may identify additional cases. These additional cases could provide a better insight in some of the aspects discussed above. Additionally, raising awareness of these rare anatomical variations amongst clinicians may lead to an additional increase in identification of these entities.

**Conclusion.** It is clear that the condition is very rare and that it rarely causes complications. However, the limited amount of research makes it difficult to make sound conclusions. Further research is required.

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#### **Conflict of interest**

The authors declare that there are no conflicts of interest

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## КРИЖОВО-КУПРИКОВІ РЕБРА ЯК РІДКІСНА УРОДЖЕНА ВАДА РОЗВИТКУ

**Джелле Станс, Аноешка Пас, Елен Девідс**

**Резюме.** Додаткові ребра – це уроджені аномалії, які можуть виявлятися в усіх відділах хребта. Найбільш рідкісним типом додаткових ребер є крижово-куприкові ребра, що розвиваються з найбільш каудальної частини хребта. Уперше крижово-куприкові ребра були описані в 1960 році і в подальшому кожні кілька років публікувалися нові одиничні дані. Іноді до опису про випадок виникнення крижово-куприкового ребра додавався короткий огляд літератури. Однак, дотепер не було опубліковано жодного вичерпного огляду літератури із аналізом етіопатогенезу крижово-куприкових ребер.

**Метою дослідження** було представити вичерпний огляд щодо додаткових ребер і зроблено спробу чітко визначити термін «крижово-куприкові ребра». Проведено порівняння з іншими типами додаткових ребер та представлено огляд літератури стосовно додаткових ребер у інших ссавців.

**Матеріали та методи.** Проведено пошук літературних даних у базах Medline та Web of Science Core Collection за кількома відповідними ключовими словами. Для визначення відповідної літератури використовувалася пошукова система Google. Також було здійснено пошук за посиланнями на публікації, ідентифіковані попередніми науковцями. Публікації були відсортовані відповідно до того, відповідають вони чи «ні» визначенню терміна «крижово-куприкові ребра». До літературного пошуку було включено всі джерела, в яких згадуються крижово-куприкові ребра, незалежно від використовуваного визначення. З огляду та аналізу літератури були виключені ті публікації, у яких не використовувався термін «крижово-куприкові ребра» та джерела, до яких не було у нас доступу.

**Результати і обговорення.** Крижово-куприкові ребра зустрічаються з однаковою частотою як у чоловіків, так і у жінок. У середньому вони діагностуються у віці 29,91 років, але цей показник коливається в широкому діапазоні (від 3 до 64 років). Такий розмах коливань крижово-куприкових ребер за віком можна пояснити тим, що такі ребра, зазвичай, виявляються випадково і присутні з народження. Крижово-куприкові ребра є рідкісною уродженою вадою розвитку і жодної інформації про фактори ризику їхнього виникнення нами не виявлено. При цьому, у більшості пацієнтів не виявлено симптомів, спричинених крижово-куприковими ребрами. Проте, деякі автори роблять припущення про те, що крижово-куприкові ребра можуть викликати певні проблеми під час пологів, що залежить від розмірів і топографії ребра, і симптоми можуть відрізнятися у різних пацієнтів.

Несвоечасну діагностику крижово-куприкових ребер можна пояснити відсутністю симптомів, пов'язаних із додатковими ребрами. Тому, для більш конкретної оцінки частоти крижово-куприкових ребер, необхідно проведення подальших досліджень. Наявні фрагментарні відомості про крижово-куприкові ребра ускладнюють чітке формулювання обґрунтованих висновків щодо етіопатогенетичних чинників виникнення цих додаткових утворень.

**Висновок.** На основі наявних літературних даних важко зробити обґрунтовані висновки щодо виникнення додаткових ребер. Проте, цілком очевидно, що така уроджена вада розвитку як крижово-куприкові ребра трапляється вкрай рідко і ускладнення, спричинені додатковими ребрами, виникають також досить рідко.

**Ключові слова:** крижово-куприкові ребра, додаткові ребра, уроджена вада розвитку.

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