

# Anestesia em Anã Acondroplásica Obesa Mórbida para Gastroplastia Redutora\*

## Anesthesia for Bariatric Surgery in an Achondroplastic Dwarf with Morbid Obesity\*

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### RESUMO

Abrão MA, Silveira VG, Barcellos CFLVA, Cosenza RCM, Carneiro JRI — Anestesia em Anã Acondroplásica Obesa Mórbida para Gastroplastia Redutora.

**JUSTIFICATIVA E OBJETIVOS:** A acondroplasia é a forma mais comum entre os diversos tipos de osteocondrodisplasias causadoras do nanismo. O anão pode ser acometido de obesidade com relativa frequência e o tratamento cirúrgico tem demonstrado maior eficácia tanto para a perda efetiva de peso quanto para a sua manutenção a longo prazo. O objetivo deste trabalho foi apresentar um caso de gastroplastia redutora com derivação intestinal em Y-de-Roux em anão acondroplásico obeso mórbido. Foram analisadas as diversas dificuldades encontradas no manejo anestésico deste paciente e a maneira pela qual foram abordadas, objetivando a diminuição da morbimortalidade no intra-operatório.

**RELATO DO CASO:** Paciente de 29 anos, feminina, anã com acondroplasia e obesidade mórbida desde a infância. Suas medidas eram de 123 cm de altura e peso corporal de 144 kg. Com índice de massa corporal (IMC) de 95,18 kg.m<sup>2</sup>, apresentava várias doenças associadas, sobretudo dos sistemas respiratório e osteoarticular. Após longo período de acompanhamento com dieta, exercícios físicos e apoio psicológico, a paciente melhorou sua condição clínica, sendo encaminhada para a realização da operação proposta: gastroplastia redutora à Capella-Fobi. Na anestesia houve dificuldade na intubação traqueal acordada sob laringoscopia direta, sendo necessária a utilização do broncofibroscópio. Transcurso intra-operatório sem complicações, sendo mantida sob anestesia geral venosa total com infusão contínua de remifentanil e propofol. Extubada ao final do procedimento na sala cirúrgica.

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**CONCLUSÕES:** As comorbidades simultâneas da acondroplasia e da obesidade mórbida podem dificultar o manejo anestésico, sobretudo em relação às vias aéreas. É necessário uma avaliação pré-anestésica bem conduzida para antecipar condutas e minimizar esses riscos, otimizando, assim, a condução da anestesia.

**Unitermos:** CIRURGIA, Abdominal: gastroplastia; DOENÇAS: nanismo acondroplásico, obesidade mórbida.

### SUMMARY

Abrão MA, Silveira VG, Barcellos CFLVA, Cosenza RCM, Carneiro JRI — Anesthesia for Bariatric Surgery in an Achondroplastic Dwarf with Morbid Obesity.

**BACKGROUND AND OBJECTIVES:** Achondroplasia is the most common form among the different types of osteochondrodysplasia that cause dwarfism. Dwarves develop obesity quite frequently, and surgical treatment has shown greater efficacy, both for effective weight loss and long term maintenance. The objective of this report was to present the case of bariatric surgery with Y-en-Roux gastric bypass in an achondroplastic dwarf with morbid obesity. The different difficulties in the anesthetic management of this patient and the way they were dealt with were discussed in order to decrease intraoperative morbidity and mortality.

**CASE REPORT:** This is a 29 years old female dwarf with achondroplasia and morbid obesity since childhood. She was 123 cm tall and weighed 144 kg at the time of admission to the Bariatric Surgery service. With a body mass index (BMI) of 95.18 kg.m<sup>2</sup>, she had several associated diseases especially of the respiratory system and osteoarticular system. After a long follow-up with diet, exercises, and psychological support, her clinical condition improved and she was referred for surgery: Y-en-Roux gastroplasty using the technique of Capella-Fobi. Intubation of the awake patient under direct laryngoscopy was difficult and a bronchofibroscope had to be used. Surgery was uneventful and the patient was maintained under total intravenous anesthesia with continuous infusion of remifentanil and propofol. She was extubated at the end of the surgery still in the operating room.

**CONCLUSIONS:** The simultaneous comorbidities of achondroplasia and morbid obesity can hinder the anesthetic management, especially regarding the airways. A thorough pre-anesthetic evaluation is necessary to anticipate the conducts and minimize risks, therefore optimizing the evolution of anesthesia.

**Key Words:** DISEASES: achondroplastic dwarfism, morbid obesity; SURGERY, Abdominal: gastroplasty.

hipóxia<sup>3,5,16</sup>. Valores preditivos de via aérea difícil, como fronte protrusa, macrocefalia, maxilar superior atrófico, mandíbula alargada, depressão do septo nasal, palato ogival, implantação anômala de dentes, pescoço curto, deformidade óssea da base do crânio, deposição de gordura cervical e apnéia obstrutiva do sono são características que se alternam ou coincidem nas duas doenças. Algumas características que alertavam para possível dificuldade de manipulação da via aérea consistiam em palato ogival, implantação anômala de dentes, deposição de gordura cervical, achatamento do septo nasal e índice Mallampati grau IV (Figuras 1 e 2). Optou-se, inicialmente, por intubação traqueal accordada sob laringoscopia direta, preservando a ventilação espontânea. Não houve dificuldade na adaptação da máscara facial durante a oxigenação prévia. A preocupação era evitar a diminuição abrupta da saturação arterial de oxigênio, por causa da baixa capacidade residual funcional e da dificuldade na manipulação da via aérea relaxada. A paciente estava agitada e ansiosa, apesar da orientação prévia sobre o procedimento e da sedação leve. É, de fato, descrita maior ansiedade nos pacientes acondroplásicos em relação à população adulta normal<sup>11,17</sup>. Houve insucesso na exposição da laringe devido à falta de cooperação e à dificuldade em girar a lâmina de um laringoscópio tradicional para a linha média, mesmo com a otimização do posicionamento preconizado em obesos mórbidos, por meio da colocação de coxins na região subescapular, ombros e cabeça<sup>5,15</sup>. A passagem de broncofibroscópio foi mais viável e tornou possível a intubação na primeira tentativa. Mayhew e col. não encontraram dificuldade de ventilação sob máscara ou intubação traqueal sob anestesia geral, em uma série de 36 procedimentos em anões acondroplásicos<sup>12</sup>. Numa análise retrospectiva de 53 procedimentos cirúrgicos ortopédicos, Moneadero e col. encontraram alguma dificuldade na ventilação sob máscara em anões acondroplásicos por causa da macroglossia. No mesmo estudo, houve insucesso de intubação traqueal em apenas um paciente em virtude da impossibilidade de visualização da laringe<sup>2</sup>. Um caso de impossibilidade total na laringoscopia direta, causada pela dificuldade na extensão da cabeça e do pescoço devido à malformação óssea da base do crânio, foi descrito por Mather e col.<sup>16</sup>. A avaliação radiológica da coluna cervical e do crânio evidencia algumas alterações sugestivas de dificuldade de intubação, como diminuição da distância atlanto-occipital, diminuição do espaço interespinhoso de C<sub>1</sub>-C<sub>2</sub> e redução da mobilidade mandibular, tornando-se, portanto, parte obrigatória da rotina pré-operatória de pacientes acondroplásicos, segundo vários autores<sup>2</sup>. Tal avaliação não foi realizada em razão da ausência de sintomas sugestivos de comprometimento neurológico, porém sua indicação é recomendável como exame de rotina mais apropriado para o estudo das displasias ósseas e como meio de avaliação de via aérea difícil.

O tamanho da cânula traqueal preconizado no nanismo é sempre menor que o tamanho convencional por causa da

hipoplasia de laringe desses pacientes e correlaciona-se melhor com o peso do que com a idade<sup>2,11,12,17</sup>. Nesse caso particular, o peso não seria parâmetro fidedigno em decorrência da desproporcionalidade resultante da obesidade. O procedimento foi realizado com duas veias periféricas de fino calibre devido à dificuldade no acesso venoso. O insucesso na venoclise tem incidência que varia entre 10% e 50% nos acondroplásicos e tem como causas o excesso de tecido gorduroso subcutâneo, a pele flácida, a deformidade articular e a ansiedade da paciente<sup>2,12</sup>. Evitou-se a punção venosa profunda, considerando o baixo risco de hemorragia nesse tipo de procedimento e o bom estado hemodinâmico da paciente. Apesar de considerada tecnicamente mais difícil em face da imprecisão das referências anatômicas, atribui-se à técnica pouco insucesso<sup>12</sup>.

A abordagem do canal espinal nos bloqueios anestésicos no neuroeixo deve ser cautelosa. As deformidades na coluna vertebral, que costumam cursar com estenose do canal, além de dificultarem a execução do bloqueio espinhal, podem causar isquemia medular, que se apresenta com sintomas neurológicos incluindo paraparesia aguda ou de progressão lenta, quadriparese, déficits sensoriais e disfunção esfincteriana<sup>1</sup>. Apesar disso, há muitos relatos de raquianestesia e peridural realizadas com sucesso em acondroplásicos, sobretudo nas cesarianas, nas quais a indicação do bloqueio oferece vantagem sobre a anestesia geral<sup>1,17</sup>. No caso relatado, evitou-se o bloqueio espinal, embora o uso de opióides por via raquidiana para a analgesia pós-operatória nas gastroplastias por laparotomia seja procedimento de rotina no serviço.

Foi possível concluir que o anão acondroplásico com obesidade mórbida deve ter avaliação pré-anestésica cuidadosa e que a escolha da conduta anestésica mais apropriada dependerá das doenças associadas apresentadas pelo paciente, bem como de suas características anatômicas.

## **Anesthesia for Bariatric Surgery in an Achondroplastic Dwarf with Morbid Obesity**

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### **INTRODUCTION**

More than 100 types of dwarfism are known and among them achondroplasia is the most common with an incidence of 1.5 in 10,000 to 1 in 40,000 live born<sup>1,2</sup>. Due to his/her physical characteristics and deformities, the achondroplastic

dwarf has several changes affecting especially the respiratory, cardiovascular, neurologic, and osteoarticular systems that are particularly related to anesthesia<sup>1</sup>. The same systems are also affected in patients with morbid obesity, leading to difficulties in the preparation and maintenance of anesthesia<sup>3-5</sup>. The association of both conditions amplifies the level of complexity, representing, therefore, a great challenge for the anesthesiologist.

#### CASE REPORT

A 29-year old female with achondroplastic dwarfism was admitted in the Bariatric Surgery Department in May 2004 weighing 144 kg and measuring 123 cm, BMI 95.18 kg.m<sup>-2</sup>. She had been obese since childhood and refractory to non-surgical treatment. Associated diseases included: sleep disorder, arthralgias with difficulty standing up and walking, intolerance to the supine position, and dyspnea on minor efforts. During 1 year and 4 months the multidisciplinary team optimized her physical and psychological condition, emphasizing supervised physical activities. When she was cleared for surgery, the patient weighed 125 kg (BMI = 82.7 kg.m<sup>-2</sup>) and her clinical improvement was evident; she was walking freely without respiratory complaints, and referred improvement in sleep quality. Her physical status was classified as ASA III and she was scheduled for vertical gastroplasty with gastrojejunostomy and silicone banding by laparotomy (Fobi-Capella). At pre-anesthetic evaluation, she had signs of bone dysplasia compatible with achondroplasia such as: shortening of the proximal segments of the upper and lower limbs and arched long bones; increased skull, mandibular and frontal protrusion, and hypoplasia of the upper jaw (Figure 1). Her neck was short, with a circumference of 38.5 cm, thyromental distance of 10 cm, and sternalmental distance of 18 cm, high palate, anomalous implantation of the incisive teeth, and Mallampati IV (Figure 2). Routine laboratorial exams were normal. Echocardiogram showed abnormal movement of the interventricular septum and ejection fraction of 0.65.

Pre-anesthetic medication included agents to prevent bronchoaspiration, such as oral ranitidine, 150 mg, and metoclopramide, 10 mg, one hour before anesthesia. Sedative drugs were not used.

In the operating room, the patient was monitored with non-invasive blood pressure, pulse oximeter, capnograph, and neurostimulator. Venoclysis was performed in the right and left upper limbs with a 22G and 20G catheter, respectively, with difficulty for the higher caliber. A kit for difficult airways was available including a flexible bronchofiberscope. Initially, awake intubation with direct laryngoscopy was attempted using a topical spray of lidocaine 10%, and bilateral upper laryngeal nerve block and transcricothyroid puncture with 2% lidocaine. Under sedation with 2 mg of midazolam and 0.01 µg.kg<sup>-1</sup>.min<sup>-1</sup> of remifentanil intravenous, three attempts of tracheal intubation were unsuccessful due to agitation and



Figure 1 – Achondroplastic Dwarf. Rizomelic Shortening of Limbs.



Figure 2 – High Palate, Anomalous Implantation of the Incisive Teeth, and Mallampati 4

patient lack of cooperation. With the aid of a bronchofiberscope, a tracheal tube with a 7-mm internal diameter was successfully inserted. Total intravenous anesthesia consisted of the continuous infusion of remifentanil 0.3 to 0.6  $\mu\text{g} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ , and propofol 75 to 100  $\mu\text{g} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ , inhalational  $\text{N}_2\text{O}/\text{O}_2$  (50/50%), and neuromuscular blockade with a total dose of 160 mg of atracurium. The intraoperative period was uneventful, with a total duration of 260 minutes for the surgery and 420 minutes for anesthesia. The patient was extubated at the end of anesthesia with the train-of-four ratio > 0.9. Intravenous dypirone 3 g, ketoprofen 100 mg, and nalbufin 10 mg, administered at regular intervals were used for postoperative analgesia. Her first complaint of pain in the first 48 hours was below three in the visual analog scale. The patient was transferred to the intensive care unit, where she stayed for 24 hours without intercurrences, and transferred to the room on the following day, being discharged from the hospital on the fourth postoperative day.

## DISCUSSION

Achondroplasia affects bone growth and it is caused by a genetic mutation of the fibroblast growth factor receptor-3 (FGFR3)<sup>2</sup>. The change in endochondral ossification causes bone malformation, which can affect primarily or secondarily other organ systems. Due to different associated diseases, the life span of the achondroplastic dwarf is 10 years shorter than the general population, and mortality is higher in the first four years of life<sup>6</sup>. Stenosis of the spinal canal and foramen magnum constitute the main cause of death in those individuals due to spinal cord ischemia leading to sudden death, especially in children younger than 4 years<sup>6,7</sup>. Obesity is a common problem in achondroplastic dwarves, with an incidence of 13 to 43%, two to eight times greater than the general population<sup>8,9</sup>. The excess body fat aggravates their comorbidities and exacerbates the difficulty in social adaptation<sup>7,8</sup>. Due to the large disproportion between weight and height it is extremely difficult to estimate the ideal weight of patients with achondroplasia<sup>9</sup>. The body mass index (BMI), which is the weight (in kilograms) over the height squared (in meters), and measurement of the triceps cutaneous fold can be used with some precision<sup>8</sup>. Other indices can overestimate the number of obese individuals among achondroplastic dwarves, which with some measurements can reach 100%. One should consider dietetic control, which should be instituted in childhood and maintained during adult life due to the early tendency for weight gain. This prophylactic measure reduces obesity in adulthood and it can improve life expectancy<sup>8</sup>. The characteristics of the musculoskeletal changes imposed by achondroplasia, with compressive neuritis, genu varum, kyphoscoliosis, and stenosis of the spinal canal affect ambulation considerably and can be associated with the greater incidence of obesity in this group of patients. Therefore, the regular practice of exercises constitutes a valuable instrument in the long term treatment of

obesity. However, the patient presented here needed a specific and personalized rehabilitation program due to her severe functional limitation, which was important to make the surgical intervention feasible<sup>9</sup>.

Half of the achondroplastic adults have measurements that are comparable to that android or abdominal obesity, which is related with greater risk for cardiovascular diseases. Several of the most severe complications of achondroplasia have to be treated as they appear or prophylactically minimizing, therefore, the impact caused by the physical incapacity and improving quality of life<sup>6,10</sup>. The main surgeries routinely done on dysfunctions caused by bone malformation include: suboccipital craniectomy and laminectomies for correction of stenosis of the foramen magnum and spinal canal, ventricular-peritoneal derivation in hydrocephalus, osteotomies for elongation of long bones and correction of tibial varus, and ENT procedure for repeated otitis media<sup>2,7,10-12</sup>. Surgery for weight loss has been widely used in the treatment of morbid obesity with good short- and long-term results. Besides, its therapeutic impact is superior to traditional treatment methods, such as diet and medication<sup>13-15</sup>. However, bariatric surgery is not widely used in achondroplastic dwarves with morbid obesity.

The morbidly obese dwarf has anomalous characteristics of both diseases frequently with synergistic deleterious effects especially in the upper airways and pulmonary function. Obstruction of upper airways and difficulty on direct laryngoscopy frequently complicate the anesthetic management of both dwarfism and morbid obesity and when associated with the impossibility of ventilation under facial mask can lead to hypoxia<sup>3,5,16</sup>. Parameters predictive of difficult intubation such as: frontal protrusion, macrocephalus, atrophic upper jaw, widened mandible, depression of the nasal septum, elevated palate, anomalous teeth implantation, short neck, osseous deformity of the base of the skull, deposits of cervical fat, and sleep apnea are characteristics that alternate or coincide in both diseases. A few characteristics that indicated possible difficult airways management had included elevated palate, anomalous teeth implantation, deposits of cervical fat, flattening of the nasal septum, and Mallampati grade IV (Figure 1 and 2). It was decided, initially, to intubate the patient awakened under direct laryngoscopy preserving spontaneous ventilation. Adaptation of the face mask during oxygenation was not difficult. The main concern was to avoid abrupt reduction in oxygen saturation, due to low functional residual capacity and the difficulty to manage the relaxed airways. The patient was agitated and anxious despite prior orientation on the procedure and light sedation. It has been described a higher level of anxiety in patients with achondroplasia than the general adult population<sup>11,17</sup>. Exposure of the larynx was unsuccessful due to lack of cooperation and difficulty to turn the laryngoscope of a traditional blade to the midline even after optimizing patient positioning with pads under the subscapular region, shoulders and head<sup>5,15</sup>. Introduction of a fiberscope was easier allowing the patient to

be intubated on the first attempt. Mayhew et al. on a series of 36 procedures in achondroplastic dwarves did not encounter ventilation difficulties using a face mask or tracheal intubation under general anesthesia<sup>12</sup>. In a retrospective analysis of 53 orthopedic surgeries, Monedero et al. found some difficulty to ventilate achondroplastic dwarves with a face mask due to macroglossia. In the same study, tracheal intubation was unsuccessful in only one patient due to the impossibility to visualize the larynx<sup>2</sup>. One case of complete impossibility to perform direct laryngoscopy caused by difficulty to extend the neck due to osseous malformation of the base of the skull was described by Mather et al.<sup>16</sup> Radiological evaluation of the cervical spine and skull show some alterations suggestive of difficult intubation such as: decreased atlanto-occipital distance, reduction of the C<sub>1</sub>-C<sub>2</sub> inter-spinous space, and reduction of mandibular mobility which is obligatory in the preoperative routine of achondroplastic patients according to some authors<sup>2</sup>. Such evaluation was not done due to the absence of symptoms suggestive of neurological compromise; however, it is recommended as the most appropriate exam to study osseous dysplasia and to evaluate difficult airways.

The size of the tracheal tube recommended in dwarfism is always smaller than the conventional size due to laryngeal hypoplasia, and has better correlation with the weight than age<sup>2,11,12,17</sup>. In the case presented here weight would not have been a reliable parameter due to the lack of proportion caused by obesity.

Two small-caliber peripheral veins were used due to the difficult venous access. The incidence of unsuccessful venoclysis in achondroplastic dwarves is between 10 and 50% and it is caused by: excess of subcutaneous tissue, flaccid skin, joint deformities, and anxiety<sup>2,12</sup>. Due to the low risk of hemorrhage in this type of surgery and the good hemodynamic state of the patient, deep venous puncture was avoided. Although it is considered technically more difficult due to the lack of precision of anatomical references, it is attributed a high success rate<sup>12</sup>.

The approach of the spinal canal in neuro-axis blocks should be careful. Spinal deformities, which generally are usually accompanied by stenosis of the spinal canal besides hindering execution of neuro-axis block can cause spinal cord ischemia that presents with neurologic symptoms, including acute or slowly progressive paraparesis, tetraparesis, sensorial deficit, and sphincter dysfunction<sup>1</sup>. But there are several reports on successful neuro-axis and epidural blocks in achondroplastic dwarves, especially in cesarean sections when this type of block offers more advantages than general anesthesia<sup>1,17</sup>. In the case reported here neuro-axis block was avoided although the use of neuro-axis opioids for post-operative analgesia in laparotomies for gastroplasties is routine in our service.

One can conclude that in achondroplastic dwarves with morbid obesity, pre-anesthetic evaluation should be thorough and the choice of the most appropriate anesthetic conduct

depends on associated diseases as well as the anatomical characteristics of the patient.

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## RESUMEN

Abrão MA, Silveira VG, Barcellos CFLVA, Cosenza RCM, Cameiro JRI — Anestesia en Enana Acondroplásica Obesa Mórbida para Gastroplastia Reductora.

**JUSTIFICATIVA Y OBJETIVOS:** La acondroplasía es la forma más común entre los diversos tipos de osteocondrodisplasias causadoras del enanismo. El enano puede ser acometido de obesidad con una relativa frecuencia, y el tratamiento quirúrgico ha demostrado una mayor eficacia, tanto para la pérdida efectiva de peso como para su mantenimiento a largo plazo. El objetivo de este trabajo fue presentar un caso de gastroplastia reductora con derivación intestinal en Y-de-Roux en enano acondroplásico obeso mórbido. Se analizaron las diversas dificultades encontradas en el manejo anestésico de este paciente y la manera a través de la cual se abordaron, objetivando la disminución de la morbi mortalidad en el intraoperatorio.

**RELATO DEL CASO:** Paciente de 29 años, femenina, enana, con acondroplasia y obesidad mórbida desde la infancia. Sus medidas eran de 123 cm de altura y peso corporal de 144 kg, al momento de la entrada en el servicio de Cirugía Bariátrica. Con un índice de masa corporal (IMC) de 95,18 kg.m<sup>-2</sup>, presentaba varias enfermedades asociadas, principalmente del sistema respiratorio y osteo-articular. Después de un largo período de acompañamiento con dieta, ejercicios físicos y apoyo psicológico, la paciente mejoró su condición clínica, siéndole intervenida con la operación propuesta: gastroplastia reductora a la Capella-Fobi. En la anestesia, hubo dificultad en la intubación traqueal con la paciente despierta y bajo

laringoscopía directa, siendo necesaria utilización del broncofibroscopio. Transcurso intraoperatorio sin complicaciones, siendo mantenida bajo anestesia general venosa total con infusión continua de remifentanil y propofol. Extubada al final del procedimiento en la sala de operaciones.

**CONCLUSIONES:** Las comorbilidades simultáneas de la acondroplasia y de la obesidad mórbida pueden dificultar el manejo anestésico, principalmente con relación a las aéreas. Es necesario una evaluación preanestésica bien conducida para anticipar conductas y minimizar esos riesgos, optimizando así la conducción de la anestesia.