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SCIENTIFIC LETTERS

Fast response of subglottic haemangiomas to propranolol☆,☆☆

Respuesta rápida de hemangiomas subglóticos a propranolol

Dear Editor:

Subglottic haemangiomas occur rarely but may be life-threatening if they become large enough to occlude the tracheal lumen. Given the difficulty of surgical access to these lesions and the high mortality they carry, the discovery of propranolol as a therapeutic agent[1] has been promising.

We conducted a retrospective study of all pediatric patients with a diagnosis of subglottic haemangioma (11 in total) treated in a tertiary level hospital between 1997 and 2014. Three of the patients (27%) were male; and all were aged between 2 weeks and 2 years (mean, 4 months). All patients were followed up until resolution of the haemangioma.

The treatment received by each patient corresponded to the customary approach applied nationwide at the time the diagnosis was made, and could be thus distributed:

Between 1997 and 2000: There were three cases diagnosed in patients aged 1, 3 and 3 months, respectively. The first patient responded well to transoral CO_2 laser microsurgery, with stridor resolving after the procedure. In the other two patients, stridor recurred a few weeks following surgery. They underwent a second CO_2 laser resection, two months after the initial surgery in the first patient, and eight months after in the second.

Between 2001 and 2009: During this period, five cases were diagnosed, with three patients receiving oral corticosteroids and two remaining under observation until full spontaneous resolution of the haemangioma. Two of the three patients that received oral corticosteroids, aged 2 and 3 months at the time of diagnosis, responded well to oral corticosteroids at doses equivalent to 1 mg/kg/day of methylprednisolone with progressive tapering off. Stridor resolved after two weeks of treatment in the first patient and after two months in the second patient. However, one patient developed iatrogenic Cushing syndrome at four months of treatment, and it was decided that she undergo CO_2 surgery in order to discontinue oral corticosteroid treatment. Of the two patients that were monitored through the spontaneous involution of the haemangioma, one had needed a tracheotomy at age 2 months due to respiratory failure, followed by the diagnosis of subglottic haemangioma, and remained under observation for one year until the lesion had resolved in full. The second case corresponded to the incidental finding of haemangioma in a patient assessed for laryngomalacia at age 2 years, who remained under observation for 10 months until the lesion had resolved spontaneously.

From 2010 on: during this period, three cases were diagnosed in patients aged 5, 3 and 2 months. All three patients responded well to treatment with oral propranolol, with resolution of stridor at three days, one week, and four days of treatment, respectively. None of the three experienced adverse effects associated with the use of beta-blockers. The lesions had become undetectable on examination by fibrolaryngoscopy at five months, one year and fifteen months, respectively.

Fig. 1 presents the treatment protocol used in patients managed with oral propranolol and marks the times at which

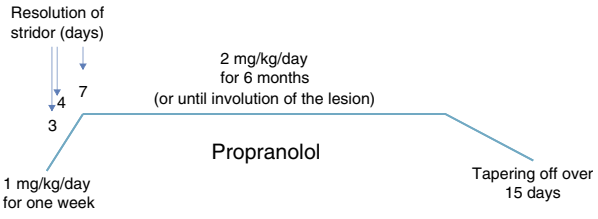

Figure 1 Oral propranolol treatment protocol used in patients with subglottic haemangioma, with the figure highlighting the rapid response of patients that receive it.

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☆☆ Previous presentations: The results of this study were presented at the 65 Congreso Nacional of the SEORL-CCC under the title *Evolución y resultados del tratamiento de hemangiomas subglóticos* (Evolution and outcomes in the treatment of subglottic haemangiomas); October 18, 2014; Madrid, Spain.

complete resolution of symptoms with absence of stridor was observed.

Our study showed good initial outcomes of CO_2 laser surgery in all patients, but also a high rate of recurrence, consistent with previous reports by other authors.[2] We also observed that when oral corticosteroids were used as the first-line treatment in a later period, patients developed complications (Cushing syndrome).[3]

However, when a propranolol treatment protocol started to be implemented in pediatric patients with a diagnosis of subglottic haemangioma, stridor resolved in our patients at three, four and seven days after initiation of treatment, so that they could be discharged to outpatient follow-up care. This quick clinical response to propranolol has been observed by other researchers.[4] The low incidence of subglottic haemangioma precludes the possibility of conducting studies with large sample sizes, and while we have observed promising results, there is not enough evidence to assert that treatment with propranolol will suffice in all cases.

To date, propranolol has allowed an effective and safe management with good disease control in the three patients treated with it. Although we have not observed any adverse effects in these three patients, we must remain alert to the potential development of adverse effects associated with beta-blockers, such as hypotension, bronchospasm and hypoglycaemia, which can be dangerous in the pediatric age group.

References

1. Léaute-Labrèze C, Dumas de la Roque E, Hubiche T, Boralevi F, Thambo J, Taïeb A. Propranolol for severe hemangiomas of infancy. N Engl J Med. 2008;358:2649–51.
2. Chatrath P, Black M, Jani P, Albert DM, Bailey CM. A review of the current management of infantile subglottic haemangioma, including a comparison of CO_2 laser therapy versus tracheostomy. Int J Pediatr Otorhinolaryngol. 2002;64:143–57.
3. Malik MA, Menon P, Rao KL, Samujh R. Effect of propranolol vs prednisolone vs propranolol with prednisolone in the management of infantile hemangioma: a randomized controlled study. J Pediatr Surg. 2013;48:2453–9.
4. Hardison SA, Dodson KM, Rhodes JL. Subglottic hemangioma treated with propranolol. Eplasty. 2014;14:ic2.

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Appendicitis in infants. 25 year case series☆

Apendicitis en lactantes. Casuística de 25 años

To the Editor:

Appendicitis has been extensively studied in early childhood (age 2–5 years), in which it is characterised by the following: (a) nonspecific and atypical symptoms (diarrhoea may be present in 33–50% of cases in this age group); (b) delayed diagnosis; (c) advanced forms of disease (gangrenous/perforated/appendiceal mass); (d) atypical bacteria and (e) a higher rate of postoperative complications.[1,2]

However, case series in the youngest age group (infants and toddlers <24 months)[3–6] are few and small in size. Our aim was to learn the particular features of appendicitis in this age group.

We conducted a retrospective study by reviewing the cases of appendicitis in patients aged 1–24 months that underwent appendectomy in a paediatric surgery department over a 25-year period (February 1990 to February 2015). To do so, we searched the medical records, narrowing them down by age and diagnosis. The analysis did not include cases of appendicitis in newborns, as they have a different aetiology and pathophysiology. We did a descriptive analysis of the following variables: clinical presentation, physical examination, diagnostic tests, duration of symptoms, findings, microbiological characteristics, and short- and long-term complications.

Thirteen young children underwent surgery, with a mean age of 20.2 months (range, 16–23). The time elapsed from the onset of symptoms to surgery was 3.8 days (range, 1–7 days). The most frequent symptoms were abdominal pain (100%), fever (92%), diarrhoea (76.9%) and vomiting (69.2%). The physical examination found nonspecific abdominal tenderness (23%), signs of peritonitis (30.8%), abdominal distension (30.8%) and omphalitis (15.4%) (Fig. 1). The workup revealed moderate leukocytosis (mean, 15,598; median, 14,092; range 9180–29,640 cells/mm^3). Ultrasound findings with identification of the appendix or appendiceal masses were only diagnostic in 23% of the patients. In all others, sonographic findings were nonspecific (mild to moderate free fluid, hypoperistalsis or lack of abnormal findings). One patient underwent computed tomography scan that led to the diagnosis of an inflammatory appendiceal mass. The surgical approach used in all cases was the classic McBurney incision. Surgery revealed gangrenous appendicitis in two patients (30.8%), perforated appendices in eight (61.4%) and appendiceal masses in three (23.1%). The bacteria involved most frequently were *E. coli* (84.6%), *P. aeruginosa* (46.1%), *E. faecalis* (46.1%), *Eikenella corrodens* (30.7%) and anaerobes (*Streptococcus milleri*, *Bacteroides fragilis*). In most cases (61.4%), more than one bacterium was isolated from the ascitic fluid. The

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