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ORIGINAL ARTICLES

Barriers and alternatives to pediatric rheumatology referrals: a survey of family doctors and pediatricians in Portugal

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ABSTRACT

Background: The access to pediatric rheumatology (PR) services in Portugal is not well described. The primary objective of this study was to identify barriers to PR referrals and explore alternative referral patterns among family doctors and pediatricians.

Methods: A web-based survey was emailed to family doctors and pediatricians practicing in Portugal to investigate issues related to access to PR care. Descriptive and comparative analyses were performed.

Results: A total of 292 responses were obtained, with 24.7% from pediatricians and 75.3% from family doctors. Only 12% claimed to have received specific education on PR. Nearly 70% worked within one hour of a PR center. Twenty-eight percent had referred a patient to PR at least once, and 9.3% considered referring to PR at least once but ultimately did not. Many referred patients to other specialties, primarily pediatrics, adult rheumatology, and pediatric orthopedics. Pediatricians encountered a greater variety of rheumatic diseases. Fifty-five percent had no opinion on the support provided by PR centers, while 24% found it sufficient. Having specific training in PR, being a pediatrician, and being a specialist were associated with higher referral rates to PR. Discrepancies in regional access to PR were documented. The most highly rated measure for improving PR referrals was promoting education.

Conclusion: The main barriers to PR referrals in Portugal are primarily the lack of education in PR, along with uneven national coverage and greater distances to some PR centers. Pediatricians appear to have better education, more experience, and higher referral rates to PR. The current alternatives for referral are pediatrics, adult rheumatology, and pediatric orthopedics. Addressing educational fragmentation was the most significant and rewarding inconsistency to overcome.

Keywords: Pediatrician; Pediatric rheumatology; Family doctor; Access to care.

KEY MESSAGES

- Portuguese family doctors (mainly) and pediatricians have a lacking education on pediatric rheumatology;
- The preferred alternatives to pediatric rheumatology referring are to pediatric, adult rheumatology and pediatric orthopedics;
- Educational consolidation was the most rated perceived enhancement approach to better referrals.

BACKGROUND

Pediatric Rheumatology (PR) is the medical specialty focused on the care and understanding of a wide range of autoimmune and auto-inflammatory diseases that affect the joints and/or have systemic effects. These include conditions such as juvenile idiopathic arthritis (JIA), connective tissue diseases (CTD), vasculitis, reactive or post-infectious arthritis, and others. Correll *et al.*[1] conducted a survey among a sample of American pediatricians, with a response rate of 15%. The study revealed that 92% of the respondents had referred patients to PR at least once during their career, while 89% had considered referring but ultimately did not. Reasons for not referring included feeling confident in managing the case independently (34%), improvement in the clinical condition while waiting (29%) or referring to another medical specialty.

The researchers observed that nearly half (48%) of the surveyed population had no specific training or education in the field of PR, and 85% had received less than four months of training. Sixty-four percent of respondents identified distance as a major barrier to referral, while 9% reported long wait times to see a pediatric rheumatologist. While data like this is available for countries as the United States of America, in Portugal, little published facts are known. While data like this is available for countries such as the

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United States of America, little published information is known about Portugal. PR has been recognized as an established medical subspecialty in Portugal since 2022, with formal recognition by the Portuguese Medical Board. Consequently, active pediatric rheumatologists in the country are primarily rheumatologists who have developed an interest and received detailed education in PR, although not within an organized subset of criteria. Therefore, there is no formal data regarding the number and distribution of pediatric rheumatologists. Some Portuguese hospitals have established specific PR clinics, but the referral patterns vary. Some clinics openly receive patients as a separate entity from Adult Rheumatology, accepting referrals from all rheumatology specialists, residents, and other medical specialties. In contrast, others receive referred patients previously sorted by a rheumatology medical supervisor. It remains to be determined whether the number and distribution of PR centers and specialists are sufficient, as well as information about waiting times, distance to PR consultations, and the level of physician education in this field. The goal of this study is to identify barriers to PR referrals and alternative referral patterns among pediatricians and family physicians, with the aim of establishing consensus on measures for improvement.

METHODS

Population of physicians being studied

All pediatricians nationwide (a solely hospital-based medical specialty in Portugal) and family doctors with a valid email address were eligible to participate in the survey. To ensure broad distribution of the survey, it was sent to high-ranking entities within both specialties, including heads of Pediatrics departments, Pediatrics residents, the Portuguese Society of Pediatrics, the Portuguese Association of Family Medicine, regional clusters of primary care centers known as ACES, and the collaborative working group between Rheumatology and Family Medicine of the Portuguese Society of Rheumatology.

Tool

The survey aimed to collect information from family doctors and pediatricians regarding their experience with pediatric rheumatic diseases, the perceived barriers to accessing PR care, and the factors influencing their decisions to refer patients for a PR consultation. The survey included questions to evaluate the respondents' experience and education in PR, including the specific rheumatic conditions they encountered and diagnosed. In order to assess barriers to accessing care, the physicians were asked about the distance between their workplace and a PR center, whether they had referred patients to a PR center previously, their reasons for referring to PR, reasons for not referring when considering it, and whether they had ever referred patients to an adult rheumatologist instead and the reasons for doing so. Additionally, the physicians were asked to grant their perception of the support provided by PR centers and were requested to rate various measures aimed at improving PR referrals. The survey included both multiple-choice and "select all that apply" questions, as well as an option for respondents to include free-text responses.

Conducting the survey

The survey was conducted from March 6, 2022, to May 10, 2022, utilizing the structure of Google Forms. The initial invitation email included not only the survey itself but also an introductory explanation of the study and a formal request for collaboration, which was distributed among the targeted population. A reminder email was sent one week before the response window closed.

Examination of answers provided

Socio-demographic and responder characteristics were analyzed using SPSSv25. Univariate analysis involved determining general frequency distributions of responses. Bivariate analysis was conducted using chi-square tests for categorical variables, and t-tests or Mann-Whitney tests for continuous variables, depending on whether the distribution was normal or non-normal, respectively.

RESULTS

We obtained a total of 292 responses. The mean age of the respondents was 35.4 years old (± SD 9.2), and 82.5% (241/292) were female. Among the respondents, 75.3% (220/292) were family doctors, while 24.7% (72/292) were pediatricians. Among the pediatricians, 61.1% (44/72) worked on secondary hospitals, and 38.9% (28/72) worked in tertiary hospitals. Overall, 55.8% (163/292) were specialized physicians, while 44.2% (129/292) were residents. Experience-wise, 34.9% (102/292) had been working as doctors for 3 to 5 years, 19.5% (57/292) for 10 to 20 years, 18.5% (54/292) for less than 2 years, 16.8% (49/292) for 6 to 10 years and 10.3% (30/292) for more than 20 years. The respondents' geographic distribution is depicted in Figure 1.

We compared family doctors and pediatricians in terms of age, gender, professional status, years of practice, and geographic setting of their workplace. Family doctors had a higher proportion of respondents who were residents (48.2% vs 31.9%, p=0.016) and respondents working for less than 2 years (21.8% vs 8.3%, p<0.001), while pediatricians had a higher

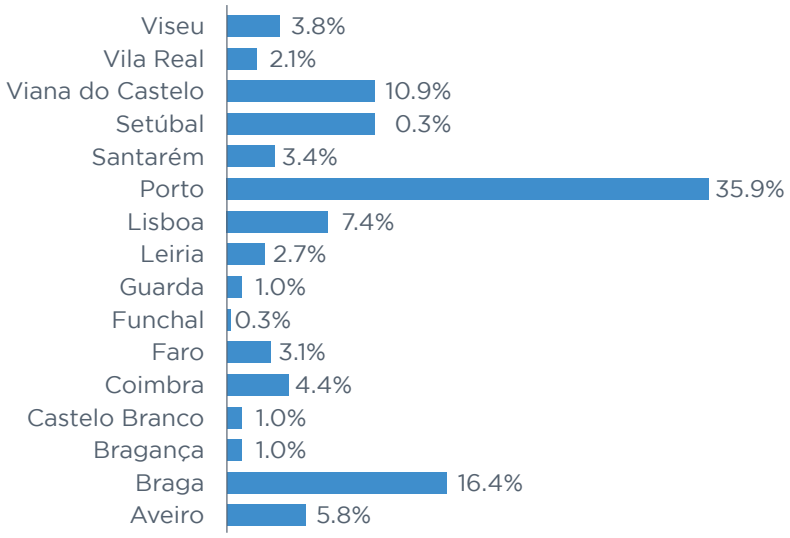

Figure 1. Respondents' geographic origin by county.

proportion of respondents working for 10 to 20 years (34.7% vs 14.5%, p<0.001). No significant differences were found between the two groups regarding age, gender, and county distribution. Further display of these results is provided in Table I.

Patterns of referral to PR

When asked if they had ever referred a patient to a pediatric rheumatologist, 28% (83/292) of surveyed physicians responded affirmatively. It is important to note that pediatricians exclusively treat children, whereas family doctors have a broader patient population. The reasons for referring patients to PR were as follows, listed in decreasing order: high suspicion for a rheumatic disease (23.6%, 69/292), unexplained fever (7.9%, 23/292), musculoskeletal pain with a normal examination (7.2%, 21/292), acute arthritis (5.8%, 17/292), positive antinuclear antibody of unclear significance (5.1%, 15/292) and chronic arthritis (4.8%, 14/292). All of these options were more likely to be selected by pediatricians as shown in Table II.

Family medicine respondents were more likely to refer pediatric patients to other medical specialties (43.6% vs 29.2%) [OR=1.157 (IC 95%: 1.019-1.316)]. The referral template is shown in Figure 2.

The most common reasons for referring patients to adult rheumatologists were considering it more convenient in terms of time/distance to access PR (42.9%, 12/28) and the patient being an adolescent (32.1%, 9/28). The analysis demonstrated a significant association between pediatrics and this type of referral due to the patient being an adolescent (p=0.001), as well as between family medicine and referral to adult rheumatologists as a shortcut to PR (p=0.001).

Obstacles to referring patients to rheumatology

The geographical proximity to PR centers as a barrier to referral was explored among the respondents. The results showed that 37% reported their workplace to be within a half to one-hour drive, 35.3% (103/292) within 30 minutes, 14.4% (41/292) between one to two hours, and 3.8% (11/292) between two to three hours away. Only 0.7% (2/292) were more than three hours away, while 8.6% (25/292) were unsure about the distance. Family medicine was associated with being two to three hours away from the nearest PR center (p<0.001), while pediatrics was associated with a time gap of less than 30 minutes (p<0.001). Respondents who perceived sufficient support from PR centers were more likely to be less than 30 minutes away (p<0.001). Referral to PR was associated with being less than one hour away from the nearest PR center (p<0.001). A total of 9% of respondents considered referring to a pediatric rheumatologist but did not, which was more common in the family medicine group [OR=1.337 (95% CI: 1.893-5.910)]. The most common reason for not referring was choosing another specialty (59.9%, 16/27).

Experience in Pediatric Rheumatology

Out of the 292 respondents, 12% reported having received specific education in Pediatric Rheumatology (PR) during their practice. Among them, 48.3% (17/35) received less than a month of training, 45.8% (16/35) had one to three months, and 5.9% (2/35) had three to five months of training. Pediatrics was statistically associated with a period of formation in PR of one to three months (p<0.001) and attendance at the most recent educational event in 2021 (p<0.001). Family doctors attended fewer

TABLE I. Sociodemographic characteristics comparison by group.

	Family Medicine (n=220)	Pediatrics (n=72)	p-value
Age (years), Mdn (IQR)	32.0 (9.0)	36.0 (13.0)	NS
Gender, feminine % (n/N)	81.4% (179/220)	86.1% (62/72)	NS
Professional status			0.016
Specialist % (n/N)	51.8% (114/220)	68.1% (49/72)	*2.4
Resident % (n/N)	41.8% (106/220)	31.9% (23/72)	*2.4
Years in practice % (n/N)			<0.001
0-2	21.8% (48/220)	8.3% (6/72)	*2.6
3-5	37.3% (82/220)	27.8% (20/72)	
6-10	17.7% (39/220)	13.9% (10/72)	
10-20	14.5% (32/220)	34.7% (25/72)	*3.7
>20	8.6% (19/220)	15.3% (11/72)	
County % (n/N)			NS
Aveiro	3.6% (8/220)	12.5% (9/72)	
Braga	16.8% (37/220)	15.3% (11/72)	
Bragança	0.5% (1/220)	2.8% (2/72)	
Castelo Branco	0.9% (2/220)	1.4% (1/72)	
Coimbra	5.0% (11/220)	2.8% (2/72)	
Faro	2.7% (6/220)	4.2% (3/72)	
Guarda	1.4% (3/220)	0% (0/72)	
Leiria	3.2% (7/220)	1.4% (18/72)	
Lisboa	7.4% (16/220)	8.3% (6/72)	
Porto	35.5% (78/220)	37.5% (27/72)	
Madeira	0% (0/220)	1.4% (1/72)	
Santarém	3.6% (8/220)	2.8% (2/72)	
Setúbal	0.5% (1/220)	0% (0/72)	
Viana do Castelo	11.5% (25/220)	9.7% (7/72)	
Vila Real	2.7% (6/220)	0% (0/72)	
Viseu	5.0% (11/220)	0% (0/72)	

NS – non-significant; Mdn - median; * adjusted residues value – positive association if > 1.96

TABLE II. Reasons for PR referral comparison by group.

Reasons for PR referral	Family Medicine (N=220)	Pediatrics (N=72)	95% confidence interval
High suspicion of rheumatic disease	18/220 (8.2%)	51/72 (70.8%)	5.10-12.05 OR=7.87
Chronic Arthritis	0/220 (0)	14/72 (19.4%)	-
Acute Arthritis	1/220 (0.5%)	16/72 (22.2%)	3.56-6.02 OR=4.63
Musculoskeletal pain with normal exam	7/220 (3.2%)	14/72 (19.4%)	2.13-4.55 OR=3.11
Positive nuclear antibody	4/220 (1.8%)	11/72 (15.3%)	2.28-4.85 OR=3.33
Unexplained fever	3/220 (1.4%)	20/72 (27.8%)	3.37-6.02 OR=4.50

OR – odds ratio; PR - pediatric rheumatology.

educational events in the last five years (median 0 vs. 1, p<0.001). Having received some form of PR training was associated with more frequent prior referrals to PR centers [OR=2.331 (95% CI: 1.623-3.344)] and was more likely due to a high suspicion of rheumatic disease [OR=2.369 (95% CI: 1.548-3.627)].

Being a specialist in their field (either pediatrics or family medicine) was also more likely to be associated with prior referrals to PR [OR=1.946 (95% CI: 1.285-2.945)]. Some regions, including Cascais, Porto, Santa Maria da Feira, Santarém, Viana do Castelo, and Vila Franca de Xira, were also statistically associated with previous referrals to PR (p=0.022).

When asked about encounters with a range of

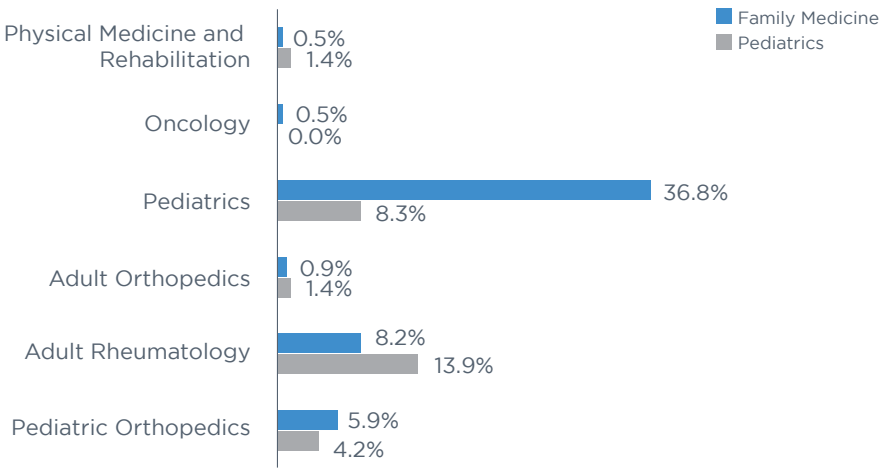

Figure 2. Other specialties' referrals.

pediatric rheumatic conditions, 47.3% (138/292) reported encountering juvenile idiopathic arthritis (JIA), 43.2% (126/292) Henoch-Schönlein purpura (HSP), 31.8% (93/292) Kawasaki's disease, 26% (76/292) systemic lupus erythematous (SLE), 9.6% (28/292) dermatomyositis (DM), 5.5% (16/292) chronic recurrent multifocal osteomyelitis (CRMO), 0.7% (2/292) periodic fever, aphthous stomatitis, pharyngitis, and adenitis (PFAPA), 0.7% (2/292) Behçet's disease, 0.7% (2/292) multisystem inflammatory syndrome in children (MIS-C), and 0.7% (2/292) amplified musculoskeletal pain syndrome (AMPS). Pediatrics respondents were more likely to encounter JIA [OR=4.629 (95% CI: 2.703-7.937)], HSP [OR=14.493 (95% CI: 6.494-32.258)], Kawasaki's disease [OR=28.57 (95% CI: 11.905-66.667)], SLE [OR=4.464 (95% CI: 3.012-6.623)], DM [OR=3.145 (95% CI: 2.183-4.525)], and CRMO [OR=4.546 (95% CI: 3.483-5.917)] compared to family doctors. Pediatricians also encountered a larger number of different rheumatic diseases (mean 3.77 vs. 1.65, p<0.001).

Perception of support provided by PR centers

When asked about their perception of the support provided by PR centers, 54.8% (160/292) of the respondents had no opinion, 24% (70/292) found it sufficient, and 21.2% (62/292) found it insufficient. Family medicine respondents were more likely to have no opinion on this topic compared to pediatricians (69.5% vs. 9.7%; p<0.001). Pediatricians were more likely to be satisfied with this support (63.9% vs. 10.9%; p<0.001), as well as those from Porto and Guimarães (p=0.008). Having received PR education was associated with a statistically significant judgement of sufficiency on this query (p<0.001).

Improving referrals to PR centers

We presented four measures for respondents to rate on a scale of 0 to 10 in terms of increasing importance to improve the quantity and quality of PR referrals. The mean rating for increasing the number of pediatric rheumatologists was 5.88 (SD 3.03), for diversifying the geographic coverage of pediatric rheumatologists was 6.50 (SD 2.96), for creating a fast-track for patients in needy areas was 6.63 (SD 2.94), and for increasing education on PR for pediatricians and family doctors was 7.24 (SD 2.99). Respondents from Braga, Faro, and Porto gave higher ratings (10, 10, and 8 respectively) for the need to expand national coverage of PR centers (p<0.001).

DISCUSSION

The majority of pediatricians responding to this survey had referred a patient to a PR center, which was not true for family doctors. This latter specialty, being broader and taking care of patients of all ages, can more easily miss some less common but nonetheless essential areas like this one. The most common overall reason for referring to PR was high suspicion of rheumatic diseases of childhood. Previous studies[1,2] found that the main reasons were also high musculoskeletal suspicion of rheumatic diseases of childhood, chronic arthritis of unknown etiology, musculoskeletal pain, abnormal lab tests and joint swelling. Contrarily to what some have reported[1], only a small minority of respondents considered referring a patient to PR but ultimately did not.

On these occasions, the survey participants redirected their referrals to other medical specialties, including pediatrics, adult rheumatology, and pediatric orthopedics. A previous study[3] showed that most children with oligoarticular JIA were referred to orthopedics before reaching PR. It would be interesting to investigate whether other specialty referrals were based on specific symptoms that caught the referring physician's attention (e.g., musculoskeletal symptoms

for orthopedics, fever for infectious diseases). Our study also found that only 12% of respondents had formal exposure to PR, and nearly half of them had less than one month of experience, particularly among family doctors. This is a significant concern for our healthcare quality and requires further improvement.

As mentioned earlier, referring patients to adult rheumatology was a common alternative. Pediatricians found it feasible when dealing with adolescent patients, and family doctors saw it as a quicker route to PR. Previous studies have shown that more than 60% of adult rheumatologists have treated pediatric patients[4], although the diseases they mainly deal with are JIA and SLE, and for patients above the age of 6[5]. While it is true that some diseases such as SLE and rheumatoid factor-positive polyarticular JIA affect both children and adults similarly, many other conditions occur exclusively in childhood and have a peculiar element to them. According to this survey, around 70% of respondents reported working within an hour's distance from a PR center.

One should exercise caution when interpreting the results, as there is a clear imbalance with a majority of respondents from the northern part of the country, and a smaller representation from more rural areas, the south and islands. Additionally, travel distance may be a more significant concern for patients and their families, which the attending physician may not fully appreciate. This study has several important limitations, including a small sample size relative to the national population of pediatricians and family doctors, differences in professional maturity between the two groups, and an unbalanced distribution of respondents across different regions. Although email is a convenient way to distribute surveys, it is also easily overlooked, despite our efforts to minimize this by sending introductory and reminder emails. Low response rates are common in surveys targeting physicians[6].

As such, the interpretation of the data from this study must be processed thoroughly, as it may not be representative of the national picture. Due to our method of propagating our query (hierarchical entities), the fact that not all of them complied with our quest, and the lack of official numbers of medical practitioners in both Pediatrics Departments and Primary Care Centers, it was very difficult to understand the total number of physicians that came in contact with this work, therefore making it impossible to calculate a response rate. It is worth noting that our survey respondents are relatively young, which could explain their higher engagement with email and greater willingness to participate compared to older individuals. A significant number of respondents, especially among family doctors, were not familiar with PR support, emphasizing the need for increased awareness and education on this matter. Furthermore, self-admittedly, increasing one's training on PR was the highest scoring measure among those offered in the survey. This supports our reasoning that academic efforts should be undertaken.

CONCLUSION

This study illustrates that pediatricians and family doctors face challenges in referring patients to PR due to uneven national PR coverage and long travel distances. Alternatively, they often refer to other specialists who may or may not have the adequate expertise to manage these diseases. These data strongly suggest the need to improve access to PR care for children with suspected rheumatic diseases. Moreover, there is a clear gap in the educational front that needs to be addressed, as the number of adequately educated physicians on PR is low, and this training was found to be associated with increased referrals and higher suspicion of rheumatic diseases. Additionally, the establishment of outreach centers or fast-track lanes, as well as the implementation of telemedicine, might theoretically be beneficial. Finally, this survey was limited by its small population (due to a combination of insufficient distribution of the survey and a low response rate), so proper generalizability of these findings requires confirmation on larger, more diverse samples that accurately represent the country's reality.

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